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Original Communications

THE CLINICAL PHYSIOLOGY OF THE HUMAN BRONCHI

III. THE EFFECT OF VAGUS SECTION ON THE COUGH REFLEX, BRONCHIAL CALIBER, AND CLEARANCE OF BRONCHIAL SECRETIONS

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A SURVEY of the available literature pertaining to the physiology of the bronchi reveals that actual experimental evidence concerning the true function of those nerves distributed to the human bronchi has not as yet sufficiently accumulated. The majority of the conclusions drawn, as well as the opinions expressed, have been based upon experimentation with various animal species. However, anatomic and physiologic correlation cannot be unequivocally made when attempting to apply these findings to man.

In a similar manner, the doubtful results obtained after applying various surgical procedures to the extrinsic pulmonary nerves of patients with intractable asthma confirm the conclusion that our knowledge of the neurophysiology of the human bronchi is at best limited. Unilateral cervical vagotomy, unilateral cervical sympathectomy, stellate ganglionectomy, and bilateral cervical sympathectomy, as well as bilateral resection of the posterior pulmonary plexus, have yielded results which are still debatable. As a consequence, we have attempted a clinical study of the function of the extrinsic nerves distributed to the bronchial tree as a means of evaluating certain surgical procedures involving these nervous pathways.

In developing this study it was decided to direct it at first toward the physiology of the tracheobronchial tree, as this might be affected by transection of one of the vagi above the origin of the pulmonary plexus, yet below that of the corresponding recurrent laryngeal nerve. In our Clinic, unilateral vagus section has been performed for a number of years in attempts to palliate patients with advanced inoperable bronchiogenic carcinoma. Clinically, we have felt that this procedure was justified, owing to the ensuing relief from cough as well as from pain.[1] Since the contralateral, apparently normal, inner-

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vated bronchial tree might be used for comparative studies, these patients presented a unique opportunity for clinical investigation. Patients in whom the extent of malignant invasion of the mediastinum suggested involvement of the sympathetic nerves or in whom the malignancy greatly distorted the main bronchial tree were excluded from this study.

Utilizing bronchoscopy, fluoroscopy, and bronchography, the following aspects of the clinical physiology of the human bronchi were studied in a group of seven patients, each of whom had one vagus trunk transected above the pulmonary plexus and immediately below the origin of the corresponding recurrent laryngeal nerve: (1) pain of bronchial origin; (2) cough reflex; (3) bronchial caliber and movements; and (4) clearance of the tracheobronchial tree.

PAIN OF BRONCHIAL ORIGIN

In previous communications[1-4] evidence has been presented which permits an evaluation of the quality and referral of pain of bronchial origin. This was obtained by stimulation of the bronchial mucosa with an electrode introduced through a bronchoscope. In general, the subsequent bronchial pain was referred to the homolateral anterior cervical region within 2 or 4 cm. of the sternum, or to the homolateral anterior cervical region within 2 cm. of the midline.[2] Following unilateral vagotomy it was demonstrated that the pain of bronchial origin was usually abolished on the homolateral side. It was consequently concluded that the homolateral vagus nerve appears to carry those afferent fibers which transmit pain from the bronchi to the central nervous system.[3]

COUGH REFLEX

It is generally accepted that the cough reflex is a protective mechanism against the inhalation of irritant substances. The impulses ordinarily responsible for a cough are set up by irritation of the sensory receptors within the pharyngeal distribution of the glossopharyngeal nerve or stimulation of the sensory endings of the vagus in the larynx, trachea, or bronchi. This reflex is characterized by a deep inspiration followed by a violent expiration.

We have studied the cough reflex in a manner similar to our investigation of bronchial pain. Following their recovery from an exploratory thoracotomy with a unilateral vagus section subsequent to the finding of an inoperable bronchiogenic carcinoma, these patients were bronchoscoped under local anesthesia which was applied only to the pharyngeal mucosa. No endotracheal anesthesia was employed. It was noted in all the patients that introduction of the bronchoscope into the main bronchus on the side of vagotomy was not accompanied by coughing, whereas introduction into the contralateral main bronchus was accompanied by violent paroxysms. It was likewise found that electric stimulation of the homolateral main bronchus produced no cough, whereas minimal stimulation of the contralateral main bronchus resulted in violent coughing.

Additional evidence that the cough reflex arising from stimulation of the homolateral bronchial tree had been abolished, although that from the contra-

lateral side had been retained, was observed during bronchography. The patients were anesthetized in the same manner as described for bronchoscopy save for the additional anesthetization of the nasal mucosa. Endotracheal anesthesia was not administered. A tracheal catheter was passed through the nasopharynx and larynx on into the tracheobronchial tree under fluoroscopic guidance. Passage, with agitation of the tracheal catheter, well into the

Fig. 1.—Bronchogram of patient H. A. taken during full inspiration six weeks after high right vagotomy. There is no appreciable difference in the caliber of the bronchi of the homolateral side as compared to that of the contralateral side. Note density of the inoperable malignancy in the right upper lobe.

depths of the homolateral bronchial tree, together with the subsequent injection of Lipiodol as well, produced no cough, whereas the same procedure performed on the contralateral bronchial tree resulted in paroxysms of severe coughing.

These results present further evidence that the vagus nerve carries the afferent fibers transmitting those bronchial impulses resulting in initiation of the cough reflex. In addition, they indicate that the afferent fibers transmit-

ting the impulses resulting in cough are carried by the homolateral nerve. There has been but little, if any, physiologic evidence that afferent innervation of the bronchial tree is derived in part from the sympathetic system.

BRONCHIAL CALIBER AND BRONCHIAL MOVEMENT

During inspiration the bronchial tree elongates while the smaller bronchi and bronchioles dilate, whereas, during expiration, the bronchial tree shortens and the smaller bronchi constrict.[5-7] In order to aerate the inexpansile por-

Fig. 2.—Bronchogram of patient H. A. taken during maximal expiration. Again there is no appreciable difference in the caliber of the bronchi of the homolateral side as compared to that of the contralateral side.

tion of lung behind and above the hilus, the lung and hilus shift forward, outward, and downward in a spiral rotary manner. Although there is some discussion as to whether or not the bronchial movements, together with changes in the bronchial caliber, are active or passive, Ellis[8] has provided satisfactory evidence that they are secondary to the respiratory movements of the thoracic wall. It has been stated that, following high vagal section in the experimental

animal, paralytic bronchial dilatation occurs.[9] Another report stated that bronchoscopic examination of the bronchial tree following unilateral cervical vagus section revealed diminished bronchial motility and excessive bronchial secretion on the homolateral side.[10]

A clinical study of the movements of the bronchial tree in our seven patients with unilateral vagotomy above the pulmonary plexus, but below the corresponding recurrent laryngeal nerve, was then made using bronchoscopy, bronchography, and fluoroscopy. In our patients so treated, using the movements and caliber of the contralateral bronchial tree as a guide, and determining the movements and caliber of the homolateral bronchial tree, valid comparisons could be made of the effects of unilateral vagotomy. These patients were examined approximately eight to twelve days postoperatively.

Bronchoscopy was performed first and the bronchial movements and caliber of the right and left bronchial trees were compared. Paralytic dilatation of the homolateral bronchi following unilateral vagotomy was not observed. There was no decreased bronchial motility on the homolateral side and no apparent difference in the amount or character of bronchial secretion between the homolateral and the contralateral bronchial trees.

Bronchography was then accomplished in order to observe the bronchial movements by fluoroscopy and to make visible records for future comparisons among our patients. Bronchograms were taken in full inspiration as well as in maximal expiration (Figs. 1 and 2).[15] Comparison of Fig. 1 with Fig. 2 discloses that paralytic dilatation of the homolateral bronchi does not occur following high unilateral vagotomy. It is also to be noted that the homolateral bronchial tree does not differ from the contralateral bronchial tree when comparison is made of the degree of dilatation of the bronchi during inspiration, as well as the degree of constriction during expiration (Table I).

It is concluded, therefore, that we are as yet unable to demonstrate appreciable differences in caliber or movement between the homolateral bronchial tree and the contralateral bronchial tree in patients who have had high unilateral vagotomy. The usual physiologic extremes of bronchial movement and caliber occur during full inspiration as well as during maximal expiration subsequent to unilateral vagus section above the origin of the pulmonary plexus. This phase of the question, however, should have further and more extensive clinical investigation.

TABLE I. THE EXPIRATORY AND INSPIRATORY DIAMETERS OF BRONCHI FOLLOWING HIGH VAGUS SECTION

PATIENT	OPERATION	CALIBER OF BRONCHI IN MILLIMETERS			
		HOMOLATERAL SIDE		CONTRALATERAL SIDE	
		INSPIR.	EXPIR.	INSPIR.	EXPIR.
H. A.	Rt. Vagotomy	7.0	5.0	7.5	6.0
P. P.	Rt. Vagotomy	6.0	5.0	5.0	4.0
A. B.	Lt. Vagotomy	3.0	2.0	3.5	2.0
M. L.	Rt. Vagotomy	4.5	3.5	4.0	2.0
D. T.	Rt. Vagotomy	3.0	2.0	3.0	2.0
R. Y.	Rt. Vagotomy	5.0	3.0	3.0	1.0
P. T.	Rt. Vagotomy	5.0	2.0	4.0	1.5
Average		4.8	3.2	4.3	2.7

Moreover, since the inception of these studies, bilateral parasympathectomy of the bronchial tree has been successfully accomplished upon three patients with intractable asthma. The results, as yet, are uncertain. It is of interest, however, that the postoperative bronchograms reveal the persistence of bronchospasm. This affords confirmatory evidence that the caliber of the bronchi is unaffected by transection of the "bronchomotor" nerve.

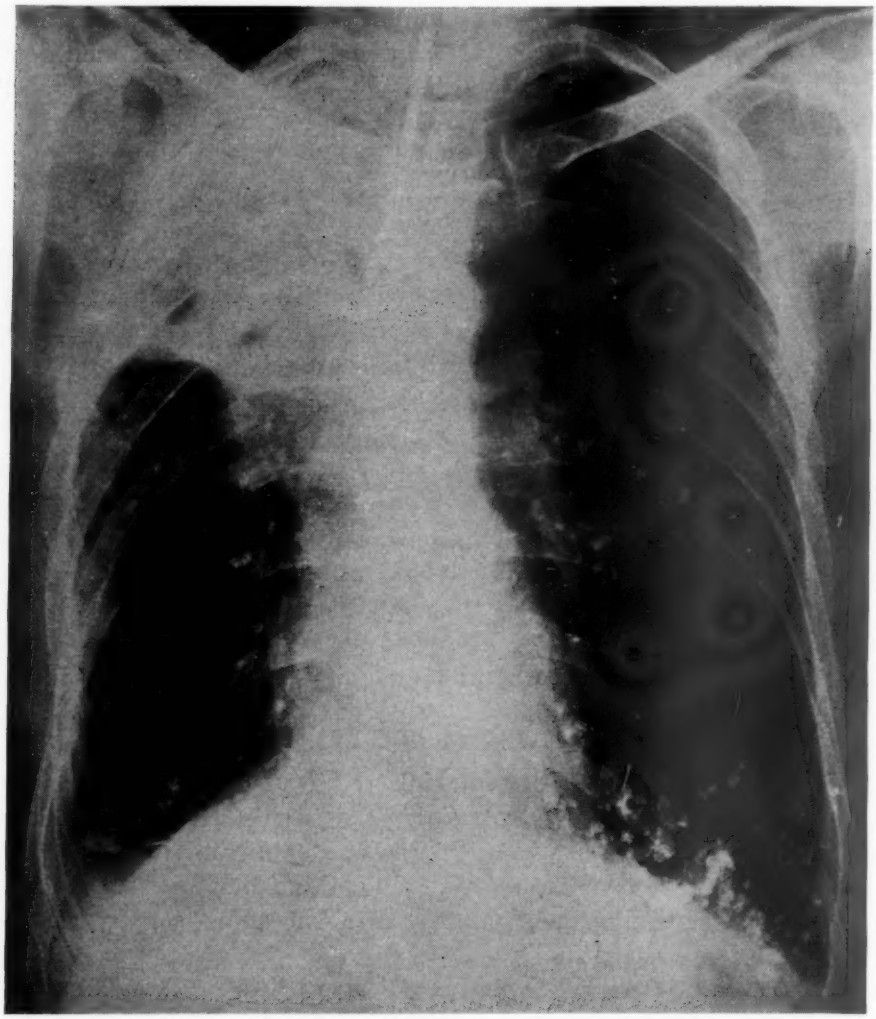

Fig. 3.—Roentgenogram of patient H. A. taken four days after the previous bronchography. Lipiodol is seen in a basal alveolar distribution bilaterally, without excessive pooling on the side of the right vagotomy.

CLEARANCE OF THE TRACHEOBRONCHIAL TREE

The tracheobronchial tree conducts air to the distal respiratory parenchyma. The patency and efficiency of the respiratory tree is maintained by three physiologic mechanisms: (1) the respiratory movements, (2) the ciliary activity in conjunction with secretions from the bronchial glands, and (3) the cough reflex.[10-14] Respiratory movements, both passive and active, are thought

to play a minor role. Theoretically, the cilia are said to be the most important factor in maintaining clearance of the smaller bronchi. Cough is regarded as most effective in the upper respiratory tree, where the gust of air may reach speeds as high as 100 miles an hour. Jackson,[11] however, believed cough as effective in the smaller bronchi, and that during the expiratory phase of coughing these smaller bronchi are compressed and the contents squeezed upwards and thus evacuated along the respiratory tree. Ciliary activity, however, requires an optimal viscosity of the bronchial glandular secretions in order to be most efficient.

In as much as vagal section abolished the cough reflex arising from the homolateral bronchial tree, a clinical postvagotomy study was made in order to compare the effectiveness of secretion clearance from the homolateral bronchial tree with the contralateral side.

Chest roentgenograms were taken one to seven days after bronchography in order to determine the residual amount of radiopaque material remaining in both sides of the bronchial tree (Fig. 3). If the residual oil in each side were found to be of approximately the same amount, then it might be assumed that unilateral vagal section had not impaired the efficiency of bronchial clearance on the homolateral side.

In six of our seven patients there was no residual pooling of secretions, such as would be evidenced by a greater residuum of oil in the homolateral side. In the one patient in whom a moderate pooling of oil was noted in the homolateral basal segments, there was a concomitant limitation of diaphragmatic movement. It was thought that this prevented complete bronchial clearance, since full activity of the diaphragm was present on the contralateral side.

SUMMARY

Seven patients, found to have inoperable bronchiogenic carcinoma at exploratory thoracotomy, afforded an opportunity to evaluate the effects of previously practiced vagotomy on the physiology of the human bronchi. Unilateral vagotomy was accomplished on these patients, above the origin of the pulmonary plexus, and immediately below the origin of the recurrent laryngeal nerve. The following results were obtained:

1. The cough reflex arising from the homolateral bronchial tree was abolished in all patients. These findings appear to indicate that the afferent fibers transmitting bronchial impulses resulting in initiation of the cough reflex are carried by the homolateral vagus nerve.

2. In the majority of our patients pain of bronchial origin was abolished on the homolateral side. In three patients there was referral of pain to the contralateral anterior cervical region. These results suggest that the afferent fibers transmitting pain from the bronchial tree are carried by the homolateral vagus nerve. The findings in the three patients in whom pain was referred to the contralateral anterior cervical region support the theory that certain of the afferent fibers from the tracheobronchial tree are carried by the contralateral vagus.

3. Bronchograms made during inspiration and expiration revealed no effects upon the normal respiratory movements of the bronchi. Paralytic dilatation of the homolateral bronchi was not found following high unilateral vagotomy. Bronchospasm persisted even after bilateral vagotomy.

4. No gross changes were noted in the amount nor in the consistency of the bronchial secretions. Likewise postbronchography chest roentgenograms revealed no impairment of the tracheobronchial clearance of Lipiodol.

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THE RELATION OF THE FACTOR OF GROWTH TO THE PATHOGENESIS OF MEGACOLON, MEGAILEUM, AND MEGADUODENUM

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SWENSON and associates[1, 2, 3] have presented evidence which supports the contention that congenital megacolon is caused by a malfunctioning and partially obstructing segment of the rectosigmoid. These authors have demonstrated that the colon returns to normal size and function after the obstruction is released.

The observations in these infants are unique, since the colonic dilatation in cases of congenital megacolon is much greater than the dilatation produced in adults by obstructing lesions of the rectosigmoid. The marked hypertrophy of adult colons proximal to obstructing lesions is well known,[4] but extremely dilated colons do not develop in adults, although they are frequently seen in infants and young children.

The fact that megacolon will develop in an infant above a rectal obstruction (Case 1) led us to perform certain experiments in puppies and in adult dogs to determine the relation of the factor of growth to dilatation of the bowel in chronic intestinal obstruction.

CASE REPORT

CASE 1.—M. A. B., a 2-day-old white male infant, was brought to Duke Hospital Dec. 27, 1942, because of an imperforate anus. He had been urinating normally and had passed no meconium through the urinary tract.

Shortly after admission a sigmoid colostomy was made, and the child got along quite well.

He was readmitted to Duke Hospital May 3, 1944, at 18 months of age, for a pull-through operation to construct a rectum. Physical examination revealed the colostomy, but the abdomen was flat and nothing else remarkable was found.

The colon was freed up and brought down to the perineum, and he began having normal bowel function.

Six months after the pull-through operation, when the patient was 2 years old, he developed a perirectal abscess and rectal fistula, with marked narrowing of the rectum. Barium enema x-ray studies at that time, however, revealed a colon of normal size and shape (Fig. 1, *A*).

Following this perirectal abscess the patient gradually developed some distention of the abdomen, although he continued to grow quite well. He was next seen on May 11, 1948, 4 years after the pull-through operation and 3½ years after the perirectal abscess. At this time he was found to have a markedly distended abdomen and a very narrow stricture of the rectum. Barium enema x-rays revealed a markedly narrowed rectum and an extremely dilated colon (Fig. 1, *B*). Rectal dilatations were carried out with moderate improvement.

This case, as well as Swenson's[1] observation of the spastic segment in patients with congenital megacolon, suggested that infants respond to obstruction of the lower colon by massive dilatation of the segment immedi-

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ately proximal to the obstructing lesion. This response in infants is quite different from that which is seen in adults, where minimal dilatation and marked hypertrophy of the musculature of the bowel are the prominent features.[4]

Even long-standing partial obstruction of the rectum as seen in lymphopathia venereum does not produce a marked dilatation of the colon. The possibility that the factor of growth may play a part in the marked dilatation of the colon was, therefore, investigated by animal experiment.

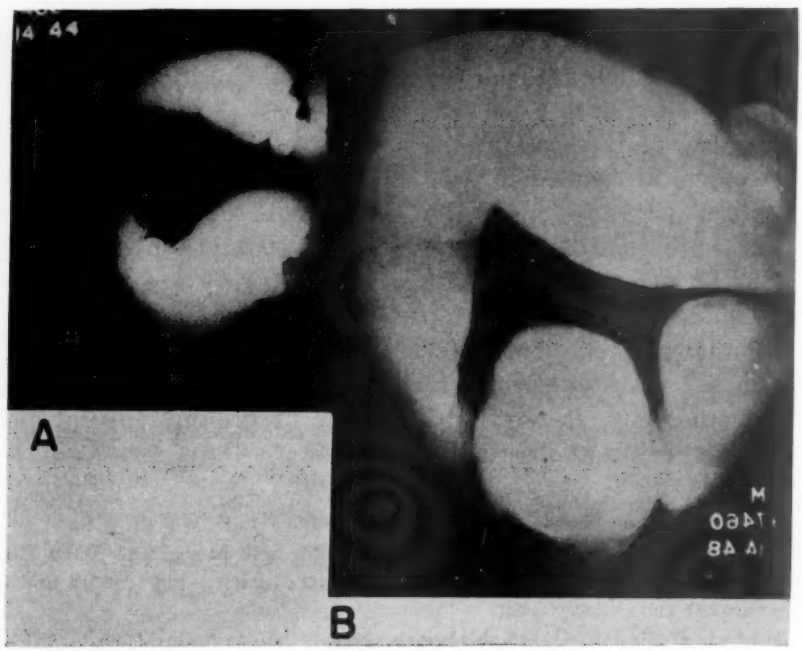

Fig. 1.—*A*, Barium enema roentgenogram of Case 1 taken shortly after the perirectal abscess. The colon was normal at that time, but the rectum was markedly narrowed. *B*, Barium enema roentgenogram of Case 1 taken 3½ years after the perirectal abscess, showing extreme dilatation of the colon, which developed in response to the obstructed rectum.

METHODS

Healthy adult mongrel dogs and newborn puppies were used for these experiments. The puppies were obtained from 6 litters which were whelped in the animal room of our surgical laboratory, and they were kept with their dames until they died or were sacrificed. The animals were prepared by fasting overnight. Intravenous Nembutal was used to anesthetize the adult dogs while ether anesthesia was given the puppies. Strict asepsis was maintained. On the adult dogs the sigmoid colon was delivered through a left lower quadrant incision. A strip of polyethylene was then placed around the sigmoid, reducing the diameter of the lumen to about 30 per cent of normal. The polyethylene was secured by tying the free ends with silk. The colon was replaced and the abdomen closed.

The puppies were prepared the same way except that the polyethylene was placed so that it was snug around the bowel, but so that it would not constrict until the size of the bowel increased with the growth of the dog. By

this means the animal could recover from the operation before he developed obstructive symptoms. The adult dogs were then fed a regular diet of commercial dog food while the diets of the puppies were supplemented with milk as soon as they were large enough to start taking the extra food.

RESULTS

After 3 to 4 months the 9 surviving adult dogs were sacrificed and the colons studied. The colons were found to be dilated up to about 1.5 to 1.7 times normal diameter, but the major response to the obstruction was a marked hypertrophy of the muscularis.

Fig. 2.—Photograph of the intestines of a puppy which had a polyethylene band around the colon for 3½ months. The arrow is pointing to the obstructing band. Note the dilated colon above and normal colon below the band.

All of the puppies died from obstruction or were sacrificed before 4 months. Fourteen of the puppies lived more than 6 weeks, however, and they were considered good experimental subjects because they had a chance to develop a chronic response to their obstructions. All of the puppies that had a chronic obstruction developed marked dilatation of the colon proximal to the obstruction. Those that died before 6 weeks were omitted because it was found that they had very little obstruction and practically no dilatation of the colon. A typical example of the dilatation of the colon which occurred in the puppies that lived more than 6 weeks is illustrated in Fig. 2.

DISCUSSION

The probability that colons of growing children respond to obstructing lesions by dilatation which is more marked than that seen in adult colons is apparently supported by animal experiments. This difference in response to

obstruction of the colon encountered in infants and children as compared to that in adults may alter the treatment of such patients occasionally, because obstruction in infants causes extreme dilatation.

Since this experimental work was done, we have encountered massive dilatation of chronically obstructed segments of small bowel in several children. Markedly dilated ileums have been seen in 2 cases in which there was partial obstruction of the terminal ileum, and marked duodenal dilatation and hypertrophy have been encountered in 2 cases in which there was chronic partial obstruction of the third portion of the duodenum (Fig. 3). We have

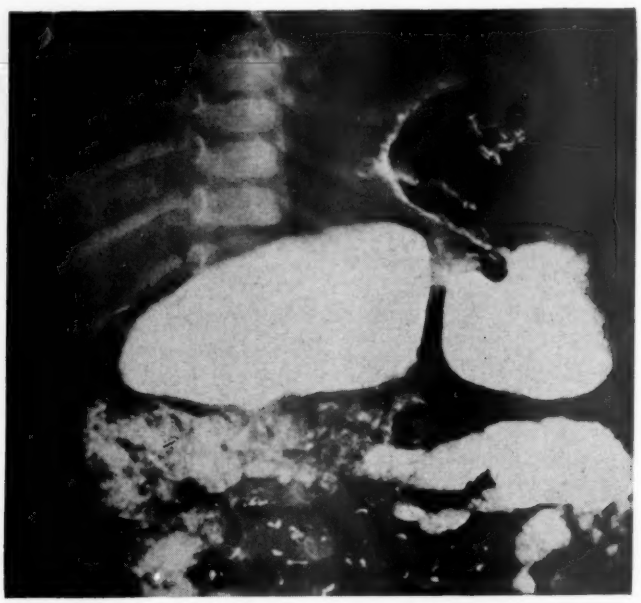

Fig. 3.—Roentgenogram with barium in the upper gastrointestinal tract of a 2-year-old child with partial obstruction of the third portion of the duodenum. The huge duodenal bulb is filled with barium.

also seen a second case which is so similar to the case reported here that a detailed report is considered superfluous; that is, the original anomaly was an imperforate anus and a stricture of the rectum producing massive dilatation of the colon, which developed postoperatively.

We feel that the dilatation is much more prominent in infants and children than in adults. This phenomenon is at present unexplained, but it is apparently related to the growth of the individual.

SUMMARY

1. A case is presented in which extreme dilatation of the colon developed in a child following obstruction to the anus.

2. Experimental data which indicate that obstruction to the colon of growing animals causes extreme dilatation of the proximal normal colon are presented and discussed.

3. The idea that partial obstruction to the colons of growing children produces extreme dilatation, while partial obstruction to the colons of adults produces predominantly hypertrophy, is proposed. This phenomenon also applies to the small bowel.

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UNUSUAL LESIONS OF THE RECTUM AND ANUS

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PHYSICIANS who perform proctologic examinations become so accustomed to finding the few common lesions of the anus and rectum that they sometimes fail to recognize certain rare pathologic changes. It would seem wise, therefore, if such lesions are not to be overlooked, to perform biopsy on every patient with a doubtful lesion and to subject to pathologic examination all surgically removed tissue from the anus and rectum, because only in this way can such lesions be recognized early enough to institute proper treatment.

During the 8 year period ending in June, 1950, proctologic examination of 30,000 private patients admitted to the Ochsner Clinic has disclosed 2 cases of rectal fibrosarcoma, 1 case of rectal leiomyosarcoma, 2 cases of malignant melanoma of the rectum, 5 cases of squamous-cell carcinoma of the anal canal, and 3 cases of rectal carcinoid tumor. Although these 13 cases represent a small percentage of the total number of patients examined, they do emphasize the necessity of looking for unusual lesions in this region. One case of each type will be briefly described and the best method of treating each type will be discussed.

FIBROSARCOMA

B. N. B., a 56-year-old white woman, was first examined in February, 1946, by her local physician because of rectal and coccygeal pain and intermittent dysentery for approximately 1 month. He discovered a pedunculated, friable, rather solid tumor which he removed from the rectum just proximal to the mucocutaneous junction in the posterior midline. The patient was referred to the Clinic for further therapy. Pathologic examination of the tumor disclosed it to be a fibrosarcoma measuring 6 by 6 by 8 cm. An abdominoperineal resection on March 30, 1946, revealed infiltration of the adnexa by the tumor. The patient convalesced rapidly and was well until a few weeks before her death in February, 1947, presumably from the sarcoma.

Comment.—Radical excision of fibrosarcomas offers the best chance of permanent cure but roentgen therapy is worth while in those patients in whom the primary tumor cannot be removed. Kallet and Saltzstein[1] reported 2 cases of patients with rectal fibrosarcomas who died 4 and 8 months, respectively, following local excision of the tumor and intensive courses of radiation. In Bacon's[2] patient who had an abdominoperineal proctosigmoidectomy because of a large pedunculated fibrosarcoma there was no evidence of recurrence 31 months postoperatively. Desjardins[3] stated that fibrosarcomas are comparatively resistant to radiotherapy but that sometimes a primary fibrosarcoma can be made to undergo more or less pronounced regression. Seldom, however, do such tumors disappear completely after radiation therapy.

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SQUAMOUS-CELL CARCINOMA

G. A. B., a 40-year-old white woman, had noted occasional loss of small amounts of bright red blood for many years, and in July, 1947, because of a tender mass protruding from the anus, consulted a physician who told her not to worry about it. In December, 1947, another physician attempted to destroy the lesion with the electric current, following which she began to have considerable pain. She came to the Clinic in March, 1948. Proctoscopic examination disclosed an ulcer 3 by 5 cm. in size on the left wall of the anal canal with several hard submucosal nodules just proximal to the dentate line on the left side. Microscopic sections of the ulcer revealed it to be squamous-cell carcinoma (grade 2). Abdominoperineal resection was done in March, 1948, and in June, 1948, bilateral inguinal node dissection was performed because of enlarged nodes in each groin. These nodes revealed only lymphoid hyperplasia. A communication from the patient in June, 1949, indicated that her health was good and that the colostomy was functioning properly.

Comment.—Many cases of squamous-cell carcinoma of the anus and anal canal have been reported. Radical excision of the primary tumor seems to be the procedure of choice.

Opinion varies regarding the indications for inguinal node dissection. Gabriel[4] believed that block dissection of the inguinal nodes should be done 2 to 3 weeks after excision of the primary tumor only if the nodes are hard and measure 1.3 cm. or more in diameter. Cattell and Williams[5] advised inguinal node extirpation in every case in which a cure is possible. Harvey[6] believed removal of inguinal nodes offers the best chance for cure. It is our opinion that radical excision of the primary tumor and bilateral inguinal node dissection should be done in all but the hopeless cases.

MALIGNANT MELANOMA

C. H. W., a 70-year-old Negro man, entered the Clinic in April, 1948, because of obstipation and low back pain which had been present for 3 months. For many years he had noted occasional loss of blood from hemorrhoids. Proctoscopy revealed a large, ulcerated, black, polypoid lesion extending superiorly from the dentate line to a level higher than could be visualized through the proctoscope. Examination of tissue obtained at biopsy disclosed a heavily pigmented malignant melanoma. Because of the considerable degree of obstruction, a loop colostomy was deemed advisable. When the peritoneal cavity was entered, myriads of black metastatic lesions could be seen throughout the entire abdomen. The patient died in April, 1949, of melanomatosis.

Comment.—As pointed out by Braastad and associates[7] in their splendid review of melano-epitheliomas of the anus and rectum, the prognosis is uniformly poor regardless of the therapy employed. They found reports of cures in only 3 instances in 94 cases; 2 of these resulted from local excision. The lesion in both of our cases was far advanced and there was no possibility of cure.

LEIOMYOSARCOMA

H. R. C., a 67-year-old white man, was admitted to the Clinic in March, 1947, because of constipation and the loss of 45 pounds during the previous 4 months. For 38 years he had had multiple draining sinuses, fistulas, and abscesses in the peri-anal region. Examination of the perineum disclosed many sinuses and a chronic ischiorectal abscess on the right side. Proctoscopy revealed a sessile polyp measuring 3 cm. in diameter at the 14 cm. level. Pathologic examination of the polyp proved it to be adenocarcinoma, grade 1. The polyp was destroyed by fulguration. When the right ischiorectal abscess was opened, it was found to contain, in addition to purulent material, a polypoid tumor measuring 6 by 6 by 8 cm. which

was attached to the rectum at the primary fistulous opening in the anal canal. The tumor was removed by severance of the pedicle but infiltration in the buttock was evident. Microscopic examination of this tumor revealed it to be a leiomyosarcoma. This patient received a course of roentgen-ray therapy after leaving the Clinic and was reasonably comfortable until 6 months before his death in August, 1948, presumably from the sarcoma.

Comment.—Few reports of rectal leiomyosarcomas have appeared in the literature. According to Rankin and Larson[8] these growths are characterized by late ulceration, rapid recurrence, and radiation-resistance and are therefore best treated by radical excision. Adelson and Newcomb[9] had a patient with an egg-shaped leiomyosarcoma which was removed by abdominoperineal resection; pathologic examination of the specimen revealed no involvement of adjacent tissue but unfortunately the patient died on the fifteenth postoperative day of probable pulmonary embolism. Bacon and Scheffler[10] reported the case of a patient who lived approximately 18 months after extensive excision of a leiomyosarcoma.

CARCINOID TUMOR

O. S., a 49-year-old white woman, was seen April 7, 1949, because of mild indigestion and fatigue. Results of all examinations were essentially negative except for a sessile, submucosal nodule measuring 3 mm. in diameter on the anterior rectal wall 4 cm. above the dentate line. This lesion was removed with the biopsy forceps and then the site was fulgurated. Pathologic examination disclosed the nodule to be a carcinoid tumor. One week later the rectal mucosa surrounding the biopsy site was excised for a distance of 2 cm. in all directions. Examination of this specimen revealed no residual tumor. Abdominoperineal resection would have been advised if neoplastic cells had been found.

Comment.—Carcinoid tumors of the rectum are rare, as pointed out by Rigdon and Fletcher,[11] who found only 14 cases reported in the literature by 1946. Pearson and Fitzgerald,[12] who recently reviewed this subject, reported that 4 of the 32 rectal carcinoid tumors reported in the literature had metastasized. They advised local excision of freely movable rectal carcinoids in young people, with frequent examinations for evidences of recurrence. More extensive lesions in elderly people demand radical excision, whereas considerable judgment is required for proper management of middle-aged patients.

DISCUSSION

It is the custom of some proctologists to fulgurate thoroughly and not to perform a biopsy in cases of rectal polypoid growths less than 3 mm. in diameter if they present no unusual characteristics. They believe this is justifiable because not only is it curative but it also saves the patient the expense of an additional laboratory procedure. Other proctologists advise biopsy of every rectal nodule before fulguration. In most instances these small polyps are found to be benign and require no treatment other than fulguration. In a few cases, however, further therapy is required, as exemplified in the patient with the carcinoid lesions. Even though biopsy and fulguration had destroyed this carcinoid tumor, we felt obligated to examine the surrounding mucosa. Destruction of these small lesions undoubtedly gives an erroneous incidence of the supposedly uncommon rectal lesions, most of which begin as polypoid growths. They probably are actually more common than the literature suggests.

SUMMARY

Thirteen cases of unusual neoplasms of the anus and rectum were discovered in 30,000 proctoscopic examinations at the Ochsner Clinic during the 8 year period ending June, 1950. These included rectal fibrosarcoma, rectal leiomyosarcoma, malignant melanoma of the rectum, squamous-cell carcinoma of the anal canal, and rectal carcinoid tumors. The importance of performing biopsy in all patients with anal and rectal lesions in order to avoid overlooking unusual lesions is emphasized.

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INTESTINAL LYMPH FLOW FOLLOWING SIMPLE INTESTINAL OBSTRUCTION IN THE RAT

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WITH THE TECHNICAL ASSISTANCE OF EMERY VAN HOOK

WANGENSTEEN[1] has discussed, on the basis of work previously done, the effects of intestinal obstruction on intestinal lymph flow. By means of excision and study of the regional lymph nodes following experimental intestinal obstruction, the absorption of vital dyes and bacteria had been determined. These studies indicated that lymphatic flow continued after obstruction of the intestine and that it was increased if there was an increase in intraluminal pressure associated with the obstruction.

The recent development of a satisfactory technique for the collection of intestinal lymph from the rat[2] offered an opportunity actually to measure intestinal lymph flow following ileal obstruction.

METHODS

The experimental animals were male albino rats of the Sprague-Dawley strain weighing 190 to 220 Gm. They were fed a standard commercial ration prior to operation. Cannulation of the intestinal lymphatic was performed by a method previously described.[2] In addition to this procedure, a linen thread was loosely knotted around the small intestine at an arbitrary level of approximately 14 inches cephalad to the ileocecal valve, or one third of the distance from the ileocecal valve to the pylorus as checked by necropsy measurement of the eviscerated intestine. The ends of the linen thread were brought out each side of the abdominal cavity through stab wounds. The last procedure was to fix a Polythene tube into the duodenum through another stab wound for the administration of 5 c.c. of 0.4 per cent solution of sodium chloride per hour by continuous slow injection. This was accomplished by a motor-driven unit in use in our laboratory. The animals were caged as previously described[3] and given nothing by mouth.

The next day, the satisfactory test animals were selected and the flow of lymph was measured hourly beginning at 8 A.M. At 11 A.M. the linen threads of alternate animals were pulled tight and the lymph flow of all animals measured every hour until 5 P.M., again at about 11 P.M., and again the following morning. The flow of lymph from one obstructed and one control animal is illustrated in Fig. 1, to show the variability of each animal. Table I, in-

TABLE I. OBSERVATIONS ON THE FLOW OF INTESTINAL LYMPH OF RATS WITH AND WITHOUT INTESTINAL OBSTRUCTION

PERIOD (HOURS)	ANIMALS WITH OBSTRUCTION		ANIMALS WITHOUT OBSTRUCTION	
	OBSERVATIONS	LYMPH FLOW (C.C. PER HOUR*)	OBSERVATIONS	LYMPH FLOW (C.C. PER HOUR*)
1-3	18	.78 ± .07	21	.70 ± .06
4-6	16	.69 ± .05	17	.63 ± .04
11-14	4	.88 ± .07	7	.83 ± .06
20-25	6	.73 ± .08	9	.61 ± .07

*The figure after the ± represents the standard error of the mean.

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cluding all the observations, shows that there is essentially no difference in the lymph flow between the obstructed and nonobstructed animals.

Necropsy of the animals revealed the ligatures to be tight and within 3 inches of the same distance from the ileocecal valve or the animal was discarded from the series. The intestines and stomachs of the obstructed animals were moderately distended with fluid above the ligature in most cases, in contrast to those of the control animals. In no case was there any evidence to suggest a significant increase in intraluminal pressure over the normal.

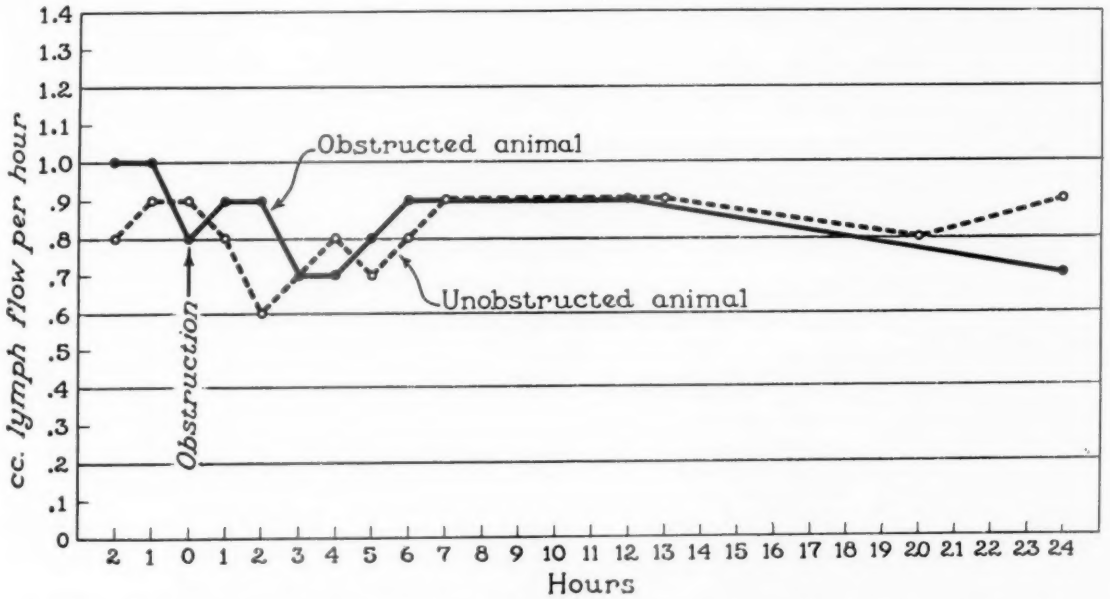

Fig. 1.—Intestinal lymph flow from one obstructed and one control animal, illustrating variability of each animal.

COMMENT

The hourly flow of lymph from the cannula of each rat often varied during the first eighteen hours of observation. This was due to intermittent plugging of the lymphatic cannula system from clotting of the lymph and removal by wire or suction. After eighteen hours the flow of lymph was usually very constant, probably because of the hypoprothrombinemia which develops.[4] The intestine was obstructed at approximately this time.

SUMMARY

Direct measurement of the flow of intestinal lymph following simple obstruction of the ileum in the rat confirmed previous deductions. The flow of lymph continued to be normal, at least in the absence of significantly increased intraluminal pressure.

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A TECHNIQUE FOR SAMPLING LYMPH IN UNANESTHETIZED DOGS BY MEANS OF AN EXTERIORIZED THORACIC DUCT-VENOUS SHUNT

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INTRODUCTION

THE purpose of this paper is to describe a new technique which makes it possible to obtain samples of thoracic duct lymph but which at the same time achieves the conservation and recirculation of the animal's lymph by employing a closed system. Continuous lymph flow into the venous system is maintained by connecting an exteriorized thoracic duct cannula to a venous catheter. Samples of lymph are easily obtained by disconnecting the tubes.

The problem of collecting lymph over a period of days has always been a difficult one. Although other investigators[1-4] have been able to collect thoracic duct lymph from some animals for 8 days, they have invariably used the technique of fistulizing the thoracic duct. This method by its very nature produces an abnormal state of body fluid equilibrium through a constant loss of lymph, and results in cachexia and general debilitation of the animals. Our technique of shunting the lymph back into the venous system permits one to take lymph samples for an indefinite period of time from an animal maintained in a relatively normal state of nutrition and fluid equilibrium. While we readily admit that there still remain inherent difficulties with the procedure herein outlined, we have received the encouragement of others because of the advantages this new approach offers. We present our findings with the hope that others in the field will be able to improve upon it and thus be able to study physiologic and pathologic changes in lymph more accurately.

MATERIALS AND PROCEDURES

Young, male, short-haired dogs weighing 35 pounds or more were used for these experiments. Surgery was performed under veterinary Nembutal anesthesia. A 10 cm. hockey stick incision was made just medial to the left external jugular vein and just superior to the left brachiocephalic vein. The proximal communicating vein,[5] which empties into the lateral aspect of the external jugular vein at a point 2 to 3 cm. above the left axillary vein, was isolated by blunt dissection and stripped. Caution must be taken not to strip the external jugular vein, as opportunity for venous thrombosis is thereby increased. The proximal communicating vein was ligated 2 cm. from its junction with the external jugular vein. Following topical application of 2 per cent procaine hydrochloride solution to relieve venous spasm, the vein was

The opinions or assertions contained herein are the private ones of the writers and are not to be construed as official or reflecting the views of the Navy Department or naval service at large.

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incised just proximal* to the ligature and a plastic catheter was inserted. The catheter was threaded into the jugular vein and down it into the left brachiocephalic vein until the tip of the catheter lay 3 to 5 cm. beyond the junction of the external jugular and axillary veins (Fig. 1).

The placement of the catheter played a prominent part in determining the success of the experiment. The flow of blood through the left brachiocephalic vein provides suction to "pull" the lymph through the tubes, thereby helping to avoid the formation of lymph clots within the thoracic duct cannula. Keeping the exposed end of the catheter at heart level, 5 to 10 c.c. of sterile normal saline solution were injected into the vein through the catheter.

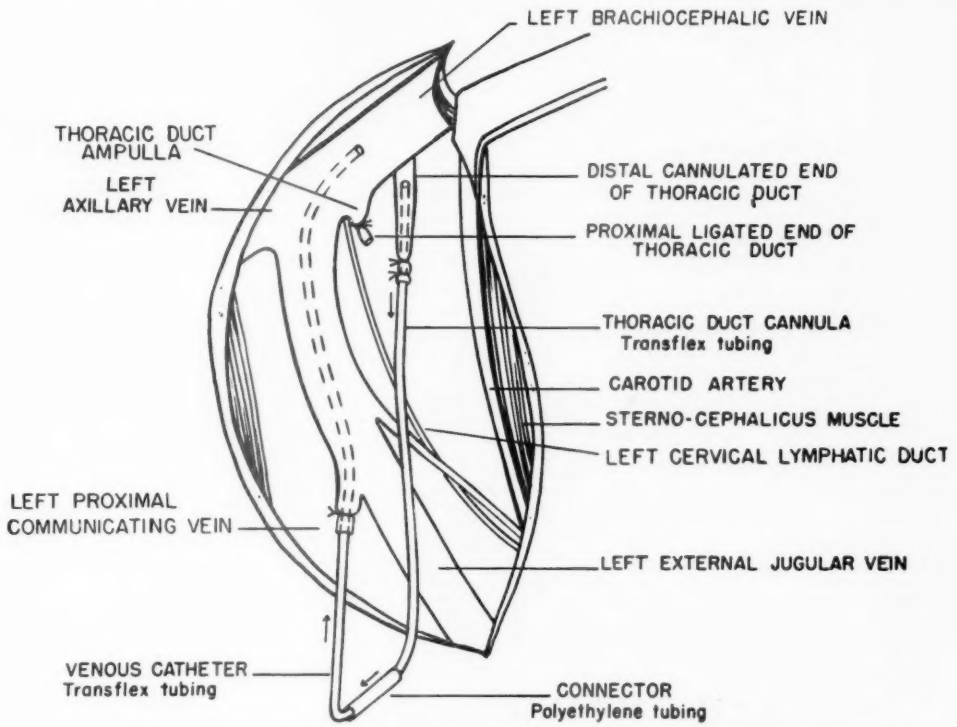

Fig. 1.—The thoracic duct-proximal communicating vein shunt in situ before closure of the incision.

The catheter was correctly placed when the saline solution was drawn into the tube and no retrograde flow of blood occurred. When the catheter lay in a satisfactory position, it was secured with a ligature placed around the proximal communicating vein as near as possible to its junction with the external jugular vein. Placing the ligature at this site diminished the possibility of the extension of thrombi from the proximal communicating vein into the jugular vein. To keep the catheter in place a ligature was tied around the catheter in the form of two half hitches, and was then sutured to adjacent muscles.

The thoracic duct is found in the fat and connective tissue lateral to the carotid artery and dorsal to the brachiocephalic vein. The duct was isolated

*The term "proximal" as used in this paper refers to that segment of vein or lymphatic nearest its termination.

by blunt dissection, but the perilymphatic tissue should be removed only at the point of cannulation. The duct was divided between ligatures placed near the ampulla. Another ligature was placed around the distal end of the thoracic duct and tied after the cannula had been inserted. The duct was incised and cannulated with a plastic tube. It is desirable to insert the cannula 4 to 5 cm. into the duct so that it will conform to the anatomic position of the duct. However, the presence of valves may prevent the cannula from being inserted this distance. The cannula was connected to the venous catheter with a short piece of polyethylene tubing (Fig. 1).

Fig. 2.—Dog with thoracic duct-venous shunt four days after operation, showing lymph flowing into collection bottle. Tube on right is venous catheter, kept closed during lymph collection with hemostat.

The wound was carefully examined for hemostasis before closure. The tissues were approximated in such a way as to provide support and avoid angulation of the cannula and the catheter. The skin was closed with 00 Dermalon sutures.

A protective covering was applied around the neck and upper thorax, allowing the animal considerable range of motion, yet preventing him from scratching the incision or pulling out the tubes. This may be done either with a plaster cast[2] or wide roll gauze between two layers of stockinette. The latter is preferable as it provides sufficient mobilization, is more easily applied, and is more comfortable for the dog. A metallic bottle-holder can be incorporated into a cast or bandage to facilitate lymph collection (Fig. 2).

The dogs were maintained on a diet of ground horse meat and Purina Laboratory Chow with water ad libitum both before and after surgery. During surgery approximately 100 c.c. of sterile 5 per cent glucose in normal saline solution were given intravenously. After surgery 300,000 units of penicillin were given prophylactically. Before application of the cast, D.D.T. powder was rubbed into the hair over the animal's neck and thorax.

Clotting of both blood and lymph is the chief source of difficulty in these experiments. In an attempt to obviate this difficulty, we used Heparin/Pitkin Menstruum.* This anticoagulant, when given subcutaneously, rapidly takes effect and remains effective in preventing clot formation for from 24 to 48 hours. The initial dose of heparin should be given shortly after surgery in order to prevent clot formation in the brachiocephalic vein. By using 3 mg. per kilogram every 48 hours, we kept the blood coagulation time as determined by the Lee-White/Howell technique[6] between 20 and 35 minutes.

As the main difficulty encountered in the majority of these experiments is clotting within the vein around the tip of the venous catheter, the cannula and catheter were inspected every 4 to 8 hours. During lymph collection periods blood can be withdrawn from the venous catheter for study. The catheter should be irrigated with sterile normal saline solution after taking each blood sample in order to insure against clotting.

A soft, flexible polyvinyl resin tubing (Transflex,† size No. 19) was used for both the thoracic duct cannula and the venous catheter. Because tissue fluids are able to dissolve the plasticizer in this tubing, it was found best not to bevel the inserted tips. A small piece of polyethylene tubing was used to join the thoracic duct cannula to the venous catheter externally.

In order to demonstrate the effectiveness of the shunt procedure, protein determinations on blood plasma and lymph were done using the micro-Kjeldahl method.[7]

RESULTS

Eleven dogs were operated upon in this series. Three dogs were operated upon but were sacrificed soon after surgery because the lymph was bloody and clot formation could not be prevented. Eight experiments were considered successful. The average length of time the shunts in these 8 animals were kept functioning was 84 hours. One dog ran for as long as 144 hours but it was necessary to replace an occluded venous catheter in this dog on the third day (Table I). In the majority of experiments the shunts ceased functioning because of clotting in the venous side. No dogs were lost from hemorrhage.

Total protein levels in both plasma and lymph remained roughly the same throughout the experiments. Table I shows the difference in portein concentrations between the first and last samples taken. Samples obtained during the intervening times fluctuated irregularly above and below these values, but consistently remained within normal limits.

*Kindly furnished through courtesy of William R. Warner & Co., New York, N. Y.

†Kindly furnished through courtesy of the manufacturer, The Irvington Varnish and Insulator Co., Irvington 11, N. J.

TABLE I. COMPARISON OF THE PROTEIN CONCENTRATIONS IN THE FIRST AND LAST SAMPLES OF BLOOD AND LYMPH TAKEN FROM DOGS WITH THORACIC DUCT-VENOUS SHUNTS

DOG	SAMPLE TIME POSTOPERATIVELY	PROTEIN				REMARKS
		PLASMA (GM./100 C.C.)	DIFFERENCE (GM./100 C.C.)	LYMPH (GM./100 C.C.)	DIFFERENCE (GM./100 C.C.)	
1	24	4.18		2.92		
	72	4.83				
	96		+0.65	3.41	+0.49	
2	0	4.30		3.76		
	96	5.16	+0.86	3.97	+0.21	Blood hemolyzed
3	0	5.72		4.69		
	48	4.25	−0.47	4.23	−0.46	
4	preoperative	4.81				Reanesthetized and new venous catheter inserted 72 hours postoperatively
	0			2.87		
	144	5.51	+0.70	3.04	+0.17	
5	preoperative	5.50				
	0			3.76		
	72	5.44	−0.06	3.20	−0.56	
6	preoperative	5.08				
	0			3.88		Animal irradiated[9] 48 hours postoperatively
	96	5.71	+0.63	3.24	−0.64	
7	preoperative	6.99				
	0			4.86		Bloody lymph at time of operation
	24	5.07	−1.92	4.71	−0.15	
8	0	4.87		3.13		Bloody lymph at time of operation
	48	5.08	+0.21	3.82	+0.69	
9	0	5.83		4.15		
	72	5.63	−0.20	4.40	+0.25	
11	0	6.30		4.63		
	48	8.13	+1.83	4.55	−0.08	Blood hemolyzed

DISCUSSION

The shunt technique provides an excellent means for studying thoracic duct lymph of animals whose fluid balance and nutritional state are almost normal. In experiments with thoracic duct lymph fistulas the animals are constantly losing protein, white blood cells, and fluids. Other investigators,[2, 8] for example, found a progressive decrease in the serum and lymph protein concentrations during their experiments. On the other hand, we found little change in protein concentration of either blood or lymph during the interval studied.

The main difficulty encountered in maintaining a lymphovenous shunt is clot formation, which can occur in either the lymphatic or venous side. We have found that several things can be done to minimize the difficulty of clot formation. First, the thoracic duct cannula should be inserted at least 2 or 3 cm. into the duct to avoid impingement of the valves or the walls of the duct against the orifice of the cannula. Second, any animals found to have bloody lymph[9] at the time of operation should be eliminated immediately as extensive clots will promptly form in the thoracic duct. Third, long-acting

Heparin/Pitkin Menstruum should be given in order to control partially the clotting at the end of the venous catheter.

From the small series of animals in which heparin was used we feel confident that long-acting heparin in proper dosage will be an important adjunct to future investigations using this technique. It is hoped that the shunt procedure will make it possible to study better the basic physiology of thoracic duct lymph in normal healthy animals.

SUMMARY

1. A technique is described for collecting samples of lymph from the thoracic ducts of dogs over a period of several days with minimal loss of lymph during this time. A lymph shunt is established by connecting a plastic thoracic duct cannula to a plastic venous catheter outside the body. Lymph flows through the tubes into the venous system when the connection is closed, or into a sampling bottle when the connection is opened.

2. Blood and/or lymph clotting is a serious complication, but can be partially remedied by using a long-acting heparin preparation (Heparin/Pitkin Menstruum).

3. This series consisted of 11 dogs. In 8 dogs functioning shunts were established and maintained for from 48 to 144 hours, with an average of 85 hours.

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PROLONGATION OF LIFE BY EMPLOYMENT OF PARENTERAL ALIMENTATION

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THE marked differences between simple intestinal obstruction and strangulation obstruction were first noted in 1892 by Kader[1] and subsequently emphasized by many investigators both experimentally[2-6] and clinically.[5, 7, 8] The results of our experiments concerning the cause of death in strangulation obstruction have been presented elsewhere.[9, 10] The portion of the work concerned with simple obstruction is herein presented.

It is of interest to note that the majority of the experimental work on simple intestinal obstruction is concerned with high obstruction,[5] a condition seen only rarely clinically.[5, 8, 11] Excellent reviews of the subject by Cooper, in 1928,[12] and Besser, in 1940,[13] have been published and no attempt is made to review the literature except insofar as is pertinent.

There is general agreement on the pathologic physiology occurring in high simple obstruction (pyloric, duodenal, or high jejunal). Death in these cases may be attributed to loss of fluid and electrolytes and the resulting dehydration. Some question, however, still exists as to the cause of death in low simple intestinal obstruction. It has been noted by many investigators that animals having a low obstruction will die usually between 8 and 14 days[14-16] without decrease in blood chlorides, hemoconcentration, or evidence of dehydration,[15] and in spite of parenteral saline solution.[5, 15] For this reason the possibility of absorption of toxic substances from the bowel lumen has never been entirely discarded.

It should be mentioned in this connection that the recent work of Abbott and associates[6] has shown that the chemical studies ordinarily taken in cases of obstruction may not necessarily reflect the true condition of the animal. There is ample evidence in both Abbott's work and the studies of Gendel and Fine[18] that a reduction of plasma volume may be of such magnitude as to cause death of the obstructed animal. Evidence against the absorption of toxic material from the obstructed gut is further afforded by the experiments of Jenkins and Beswick,[19] who have kept dogs alive for as long as 70 to 80 days by feeding electrolytes and nutritive material through an enterostomy below either a high or moderately low obstruction. No cause for death other than the long-standing inanition and its resulting complications could be found.

That the mechanical factor of distention and not a "toxic factor" accounted for the lethal issue in ileal obstructions was indicated by Wangensteen and Rea in 1939.[20] By occluding the esophagus and thereby excluding

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swallowed air, dogs having an ileal obstruction were maintained for 19 to 57 days with an average survival time of 36 days. Large amounts of parenteral saline solution and occasional blood transfusions were administered. At necropsy the gut was usually found to be collapsed, and it was felt that death was due to starvation. Their experiments indicated that if the distention factor with its attendant sequelae of altered permeability and decreased viability could be obviated, then simple ileal obstruction could be well tolerated for days and that when death did occur it was not due to any toxic factor.

We have attempted to obviate the factors of electrolyte imbalance, loss of circulating volume, and inanition as a cause of death in animals having a low simple obstruction by the parenteral administration of saline, glucose, Amigen,* blood, plasma, and gelatin and to study the course of animals so treated. We felt that it would be of interest to see how long obstructed dogs could be maintained on parenteral alimentation, and to determine whether at the same time further information on the cause of death could be obtained.

METHOD

Ten healthy adult dogs weighing between 10.6 and 15.0 kilograms, which were observed in the colony for 10 days or longer, were used in this experiment. Blood chemical studies were taken on the day before or the morning of operation. All animals were starved for 24 hours before operation except Dog 2, which inadvertently received food within 12 hours of operation. Operation was performed with the animal under veterinary Nembutal anesthesia (24 mg. per kilogram) using strict aseptic technique throughout. The abdomen was opened through a right rectus incision, and at a point 100 to 150 cm. from the ligament of Treitz (depending on the size of the animal) the bowel was severed and both ends closed by the Parker-Kerr technique. Following the occurrence of an intussusception with gangrene and necrosis of the distal turned-in end in 2 of the animals (Dogs 4 and 6), steps were subsequently taken to prevent this by suturing the end of the distal turned-in end to the side of the proximal loop. Using this technique, intussusception was avoided in the remaining animals. The abdomen was closed in layers and the operation was usually completed in around 1 hour.

The following studies were done at various intervals throughout the course of survival: blood and plasma specific gravities, plasma protein, hemoglobin, hematocrit,[21] chloride,[22] urea nitrogen,[23] amylase,[24] lipase,[25] and total nitrogen[26] of the vomitus. All animals were autopsied immediately after death.

Postoperatively, fluids were administered daily either by drip intravenously with the animal on the table, or, in the later experiments, by constant intravenous drip through a capillary plastic tube (polyethylene) threaded into the jugular vein or inferior vena cava after the method of Rhode, Parkins, and Vars,[27] except that a DeBakey pump was not used. The amounts of fluids and calories to be administered were determined by the output (urine and vomitus) and chemical studies of the blood. However, it was not always pos-

*Mead Johnson & Company, Evansville, Ind.

sible to meet these requirements because of the limitations of the methods used for administration; namely, the length of time the animal would remain on the table, thrombosis of veins, dislodgement of the plastic tube, and inconstancy of rate of flow. Water and dog biscuits were placed in the cages of the animals receiving fluids intermittently but not in the cages of those receiving constant parenteral alimentation. The dog biscuits were seldom, if ever eaten. One exception to this procedure was Dog 10. For the first 10 days after obstruction, due to much technical difficulty with the plastic tubing, an average of 140 c.c. of milk daily was allowed and given orally. Thereafter, nothing was allowed by mouth except 10 c.c. of a vitamin mixture daily which was given in addition to the parenteral vitamins.

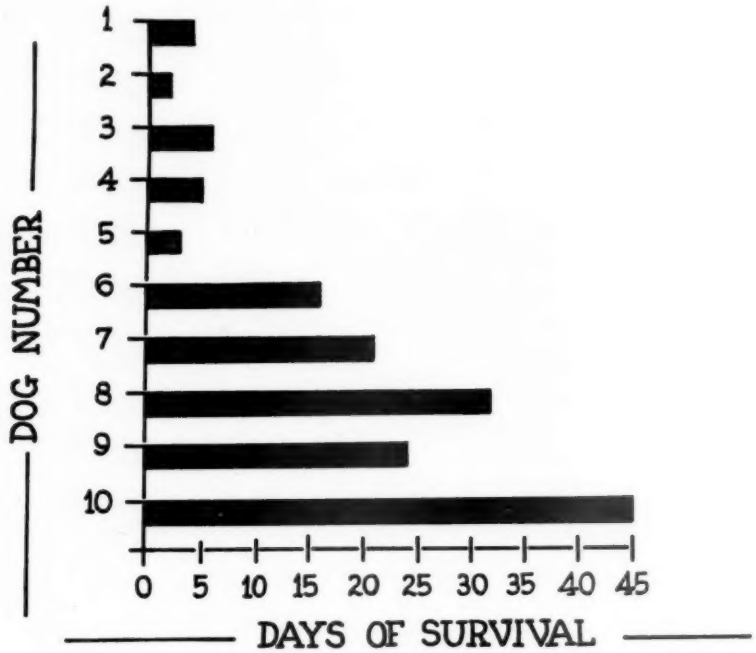

Fig. 1.—Length of survival of 10 dogs having a simple intestinal obstruction. The first 8 animals received parenteral fluids by intermittent intravenous drip. The last 2 animals received intravenous fluids constantly during the period of survival.

RESULTS AND DISCUSSION

Five of the 10 animals died between 2 and 6 days (Fig. 1), 2 of electrolyte imbalance, 1 of intussusception with gangrene and necrosis of the distal turned-in end, 1 of leakage at the suture line with a resulting peritonitis, and 1 of distemper, with complete consolidation of all but one lobe of the lungs.

The remaining 5 animals lived between 16 and 45 days, with an average survival time of 27.6 days. One died suddenly at 16 days and necropsy revealed an intussusception of the distal turned-in end with gangrene and necrosis of 8 inches of the bowel. Another was reoperated upon at 21 days, while in good condition, and the intestinal continuity was restored. The remaining 3 dogs survived for 24, 32, and 45 days, respectively. This report is primarily concerned with this group of animals.

Dogs 6, 7, and 8 (Table I) received fluids by intermittent intravenous drip. We were seldom able to administer more than a total of 500 c.c. of the various fluids intravenously in a 24 hour period. It was not possible to keep an accurate account of the output, for the animals would frequently vomit large amounts on the table while receiving Amigen or plasma or blood. If the rate of flow of the Amigen was slowed sufficiently to prevent the vomiting, then only about 100 c.c. could be delivered before the animal became restless. It will be noted (Table I) that the dogs lost between 20 per cent and 36 per cent of the body weight during the period of obstruction. The caloric intake in these 3 animals was markedly inadequate to maintain nutrition, never averaging over 11 calories per kilogram or roughly one-third of the amount we wished to obtain. Despite frequent small blood transfusions, the hematocrit and hemoglobin values markedly decreased. The hematocrit decreased from 46 to 22 and the hemoglobin from 16.3 to 8.0 in the animal which lived for 32 days.

It was apparent that adequate parenteral alimentation could not be approached by intermittent administration, and the continuous infusion method was therefore instituted in Dogs 9 and 10. These 2 animals lived for 24 and 45 days, respectively. As these animals were kept in metabolism cages while receiving fluids (except for blood and plasma, which were given on the table) accurate output data could be obtained.

TABLE I. LENGTH OF SURVIVAL AND AVERAGE DAILY FLUID AND CALORIC INTAKE OF DOGS 6 THROUGH 10*

DOG	LENGTH OF SURVIVAL (DAYS)	DAILY AVERAGE						
		INTAKE (C.C.)	CHO (GM./KG.)	N (GM./KG.)	CALORIES (KG.)	NaCl (GM.)	OUTPUT	PER CENT WEIGHT LOSS
6	16	425	1.93	0.03	7.91	3.19		30.8
7	21	450	1.83	0.02	7.42	3.67		20.0
8	32	432	2.36	0.06	10.36	2.97		36.7
9	24	1,254	8.85	0.25	39.64	8.24	1,001	18.0
10	45	1,091	6.30	0 23	29.95	6 24	722	26.0

*The output in Dogs 6, 7, and 8 could not be accurately determined. Note the difference in achieving adequate caloric requirements in the dogs receiving intermittent parenteral fluids as against the 2 receiving constant intravenous alimentation.

Both animals were maintained in satisfactory electrolyte balance throughout the course of survival. The hematocrit, hemoglobin, and serum protein were maintained in the animal which died at 24 days, but anemia and hypoproteinemia became marked around the fifth and sixth weeks in the dog which died at 45 days, despite a total of 850 c.c. of whole blood and 300 c.c. of plasma during the last 8 days. Both dogs vomited large amounts of fluid. Dog 9 vomited on 15 of the 24 days, frequently 1,000 to 1,500 c.c. or more. Dog 10, on the other hand, vomited only on 13 of the 45 days, usually at 3 to 4 day intervals. Further reference will be made to this. The animals became gradually weaker. Dog 10 was able to walk and even run about up until the forty-second day, but it arose with difficulty thereafter. Dog 9 remained in good condition up until the twenty-fourth day, when he was found moribund.

Autopsy findings in the 3 cases were essentially similar except for the findings in the veins, subsequently mentioned. Emaciation was marked. In all cases the peritoneum was smooth and glistening. There was no free fluid in the abdomen and no evidence of peritonitis. The obstructed segments of bowel were enormously distended, shortened as much as 8 to 12 cm., and markedly hypertrophied. There was in no case any evidence of strangulation or compromise of the blood supply.

The fluid content of the obstructed segments ranged from 200 to 475 c.c. When one considers this large amount of fluid collecting in a relatively short segment of the dog's gut, the large fluid loss occurring into the lumen of the gut in a patient with intestinal obstruction or paralytic ileus thus becomes evident.

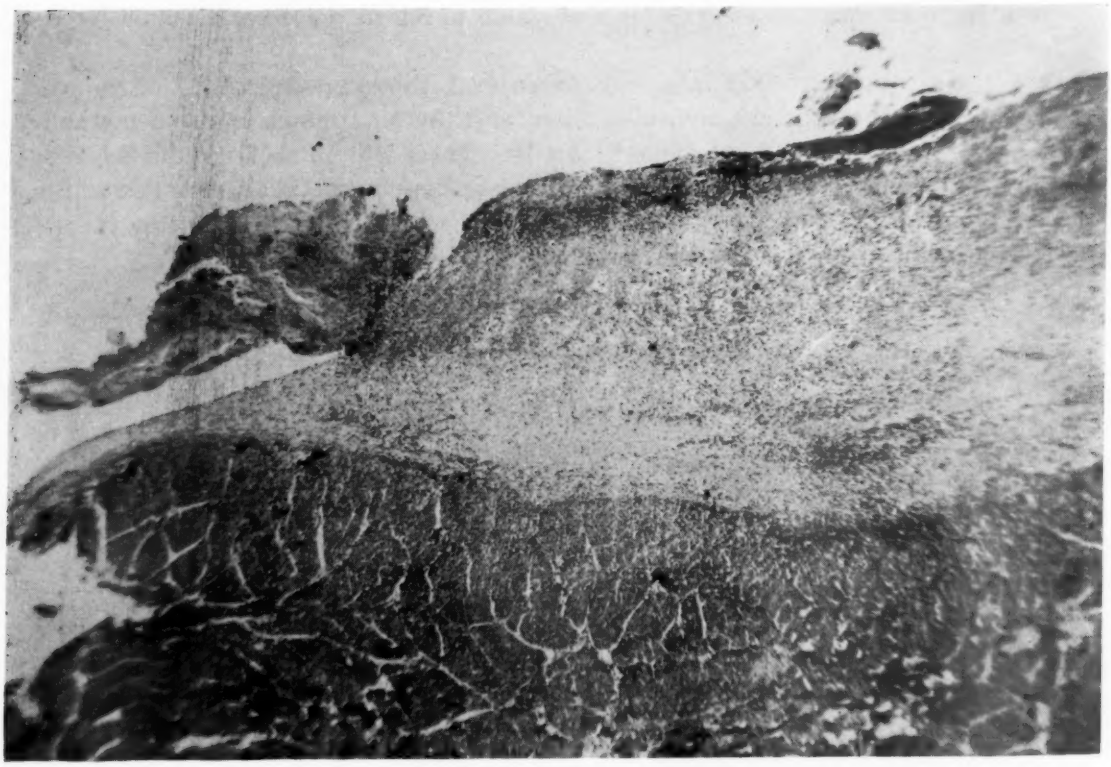

Fig. 2.—Photomicrograph showing auricular vegetations in Dog 10. The section was taken through a large vegetation on the anterior wall of the auricle (×34).

As stated, the amount and types of fluid which were administered were theoretically calculated on the basis of output (urine and vomitus), the chemical determinations on the blood, and the minimum requirements of calories and nitrogen calculated to maintain a patient in nitrogen balance[28] after operation: namely, 30 calories per kilogram per 24 hours, and 0.3 Gm. of nitrogen per kilogram per 24 hours. Actually, this was not attainable by the methods used. Mechanical difficulties and thrombosis of veins were previously mentioned as limiting factors. Another source of difficulty in caloric replace-

ment was the high temperature elevations that occurred in the 2 animals which received constant intravenous fluids. The temperature averaged between 102.4° F. and 103° F. daily for one of the dogs, and between 102.2° F. and 102.7° F. for the other. The cause for the temperature elevation was probably due to the method of delivering the fluids. Thrombosis secondary to venous catheterization with plastic tubing in the 2 animals is considered in detail in another communication,[29] but brief mention of these animals is pertinent.

In Dog 9 the polythene tube was passed for a ditance of 24 cm. into the left jugular vein and was dislodged after 120 hours. The animal received between 700 and 1,900 c.c. of the various fluids in each 24 hour period. The tube was then passed into the right jugular vein for 21 cm. and remained in situ for 19 days, until the death of the dog. At autopsy the right jugular vein was completely thrombosed from the point of entry of the tube to the superior vena cava. There was a small vegetation 0.2 by 1.0 by 0.3 cm. in the superior vena cava just opposite the right jugular orifice. At the junction of the superior vena cava and auricle there was a round, large, ragged vegetation 1.5 by 2.0 cm. on the posterior wall. The tube was resting in this vegetation. The vegetation was wide based and firmly attached at all points.

In Dog 10 the tube was passed into the left jugular vein and dislodged after 24 hours, then into the right jugular vein and dislodged after 24 hours, then back into the left jugular and dislodged after 48 hours, into the right foreleg vein for 72 hours, then the left foreleg vein for 96 hours, into the right femoral vein (into the inferior vena cava) for 24 hours, then into the left femoral vein for 19 days, and finally into the right jugular vein for the last 10 days. The difficulty which we had with this animal is thus apparent. At autopsy, both jugular veins were completely thrombosed from the entrance of the tube to the superior vena cava with a firm, white, fibrous clot. There were vegetations and ulcerations in the superior vena cava opposite the right jugular orifice. At the entrance of the superior vena cava into the auricle, on the anterior wall, there was a large vegetation 0.75 by 0.5 by 0.5 cm. This had a cauliflower appearance (Fig. 2) and extended well into the lumen from a broad base. Both iliac vessels were completely thrombosed for a distance of approximately 9 cm. The inferior vena cava was thrombosed for a distance of 10 cm. from the iliac bifurcation. At the craniad end of the thrombosis there was a firmly adherent vegetation, about 3.0 by 2.0 by 2.0 mm. floating in the lumen. The vessel wall was smooth above this point. The left leg of this animal became edematous on the thirty-first postoperative day and the edema increased to a maximum at the thirty-ninth day. Large collateral venous channels developed over the anterior abdominal wall.

An analysis of Table II shows that only in the last 14 days in Dog 9, and in the last 15 days in Dog 10, were we able to reach the theoretic goal for

TABLE II. INTAKE AND OUTPUT DATA, WEIGHT LOSS, AND TEMPERATURE BY PERIODS IN DOGS 9 AND 10*

		DAILY AVERAGE								
								WEIGHT LOSS		
DOG	PERIODS	INTAKE (C.C.)	CHO (GM./KG.)	N (GM./KG.)	CALORIES (PER KG.)	NaCl (GM.)	OUTPUT (C.C.)	(KG.)	PER CENT	TEMPERATURE (F.)
9	First 10 days	1,155	7.35	0.19	32.63	7.54	893	0.10	0.8	102.2
	Last 14 days	1,324	9.92	0.30	44.66	8.73	1,079	0.14	1.0	102.7
10	First 10 days	881	4.36	0.14	22.45†	4.81	396	0.06	0.4	102.4
	Second 10 days	957	5.15	0.18	23.86	5.58	745	0.12	0.8	103
	Third 10 days	1,230	7.20	0.23	32.68	7.55	834	0.07	0.5	102.6
	Last 15 days	1,226	7.76	0.33	37.20	6.78	850	0.09	0.7	103

*Note that only in the last 14 days in Dog 9, and in the last 15 days in Dog 10, did nitrogen intake reach that theoretically calculated to maintain the animal in nitrogen balance. Despite this, the average weight loss increased.

†Includes calories from fat in milk administered for first 10 days.

caloric requirements. Even this, in the face of a daily temperature elevation from 102° F. to 103° F., and the vomiting of large amounts of fluid, was not sufficient to prevent a weight loss of 0.7 to 1.0 per cent of the total body weight per day. The weight loss was similar to that sustained by the dogs in the experiments of Jenkins and Beswick,[19] which were fed through an enterostomy tube below the obstruction, and somewhat less than the dogs in the experiments of Kunde,[30] which were simply starved from 37 to 41 days.

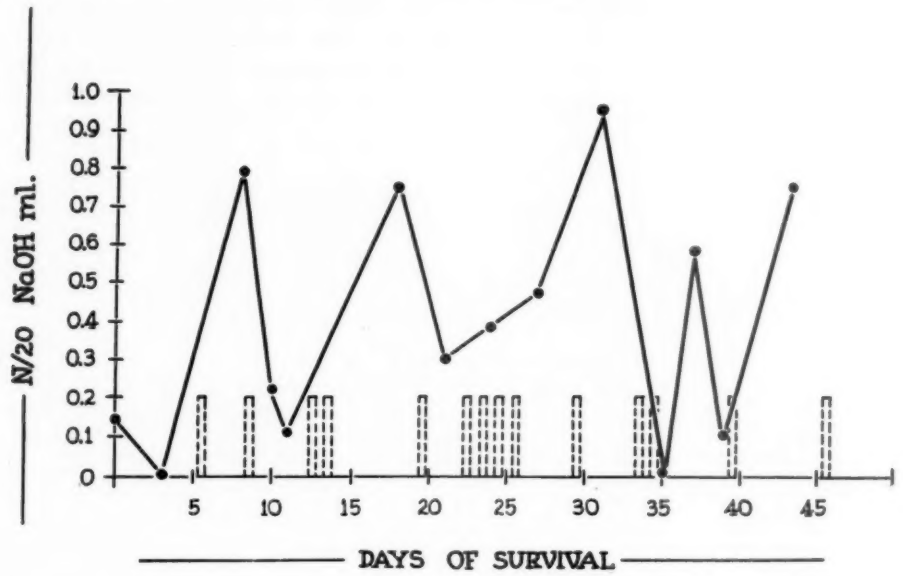

Fig. 3.—Serial serum lipase determinations in Dog 10, obstructed for 45 days. The dotted columns represent the days on which the animal vomited large amounts of fluids. Note that the increases in serum lipase occurred at a time when the animal had not vomited for several days.

The urine and vomitus were analyzed for total nitrogen on numerous occasions and were usually found to be almost twice the magnitude of the nitrogen intake. Increase in the serum lipase of animals having experimentally induced appendical peritonitis was previously reported.[31] Serial determinations were performed in this group of animals also. In 3 of the animals significant elevations of the serum lipase occurred. Bockus[32] reported increases of the serum lipase, the cause of which was not determined, in 4 cases of intestinal obstruction. An analysis of serial serum lipase determinations in the animal which lived for 45 days suggests the cause for this increase. As stated, the animal would go for several days without vomiting and then would vomit copious amounts. It will be noted (Fig. 3) that serum lipase increases occurred during those times when the dog was markedly distended and had not vomited for several days. Once vomiting occurred, the serum lipase rapidly fell. The other 2 dogs showing an increase (1.01 and 1.28 c.c. respectively at the third postoperative day) were also distended at this time. It would appear that as the obstructed gut distended, external pressure was exerted on the pancreas, thereby causing an increase in the lipase, or else that the increase

was due to the mechanical blockage of the duct orifices by the large amounts of fluid in the gut lumen. The animal which died at 24 days vomited on 15 of the 24 days and serial serum lipase determinations at around 3 day intervals were never over 0.33 c.c.

It should be stressed that in none of the animals that died of intestinal obstruction was there any evidence of a "toxemia." Certainly there was nothing comparable to the rapid and sudden demise of the animals having a strangulation obstruction,[9] nor was there any pathologic evidence of a similar process. None of the animals except the 2 with intussusception had free peritoneal fluid, and not even in these cases did the fluid resemble that noted in the strangulations. In the 3 dogs in which other causes of death could be ruled out, such as electrolyte imbalance, pneumonia, and intussusception and gangrene, the demise of the animal was related to inanition or complications resulting therefrom. In 2 of the animals in which caloric requirements were more adequately fulfilled, the length of survival was 24 and 45 days, respectively, and, despite high daily temperature elevations and losses of copious amounts of fluid by vomiting, the weight loss was of smaller magnitude than in the animals less adequately treated.

CONCLUSIONS

1. There is no evidence of a toxemia of intestinal obstruction in animals having a low simple obstruction.

2. As long as strangulation does not supervene, the length of survival of dogs having a low simple obstruction depends upon the ability to maintain fluid, electrolyte, and nutritional requirements.

3. By parenteral alimentation with the methods used adequate nutritional requirements were not obtained, but in one animal in which these requirements were approached more closely, the length of survival was 45 days.

4. Increases of the serum lipase may occur in intestinal obstruction and it is apparently due to mechanical pressure on the pancreas by the distended gut, or blockage of the pancreatic duct orifices by the large amount of intralumenal fluid.

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THE ADRENAL CORTEX AND POSTOPERATIVE GASTRO-INTESTINAL SECRETIONS

Clinical Significance of These Effects

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THE literature contains few quantitative reports on the normal daily volumes of the various components of intestinal fluid in man. This is due largely to the difficulties encountered in measuring these secretions. It was stated by Gamble that the total volume secreted into the intestine each 24 hours is in excess of 8 liters.[2] In disease states, however, the amount of fluid poured into the bowel may vary enormously. A knowledge of these variations is of basic importance to the surgeon.

As an incidental finding in a study of the effect of changes in adrenocortical activity on the postoperative urinary excretion of water and electrolytes[4] it was noted that the volume of intestinal contents aspirated through the Miller-Abbott tube in patients subjected to colon resection was decreased immediately following operation. This low volume of aspirated intestinal contents was accompanied by a decreased urinary volume and an increase in adrenocortical activity, as indicated by a profound fall in the total eosinophil count.[5] During the period when adrenocortical function was maximal, the volume of aspirated intestinal fluid and the volume of urine secreted were diminished. Conversely, as the overactivity of the adrenal cortex began to subside, as indicated by a rise in the total eosinophil count, the volume of urine and of intestinal contents increased. The records of 29 additional patients subjected to colon resection have been studied. Ten patients subjected to choledochostomy were also studied for the purpose of learning whether the output of bile through the T tube is significantly decreased immediately following operation. These results form the basis of the present report.

PROCEDURE AND RESULTS

The clinical records of 29 patients having colon resection, hereinafter referred to as Group 1, were analyzed as to fluid intake, urinary volume, and the volume of intestinal contents aspirated with the Miller-Abbott tube during the first three 24 hour periods postoperatively. The cumulative data are listed in Table I, and are presented graphically in Fig. 1. Three patients were investigated with a view to correlating the volumes of aspirated intestinal contents with the total eosinophil counts during the first three 24 hour periods postoperatively. These results are shown graphically in Fig. 2.

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The average volume of bile that drained from the T tube in the 10 patients subjected to choledochostomy varied only slightly during the first three 24 hour periods postoperatively, being approximately 200 to 300 ml. daily.

DISCUSSION OF RESULTS

The results presented in Table I and Fig. 1 confirm the previous impression that the volume of intestinal fluid which can be aspirated by Miller-Abbott tube and continuous suction is decreased immediately following the trauma of anesthesia and operation. Furthermore, in Fig. 2 it may be seen that the duration of this diminished volume of intestinal secretion is concomitant with an increased activity of the adrenal cortex, as evidenced by

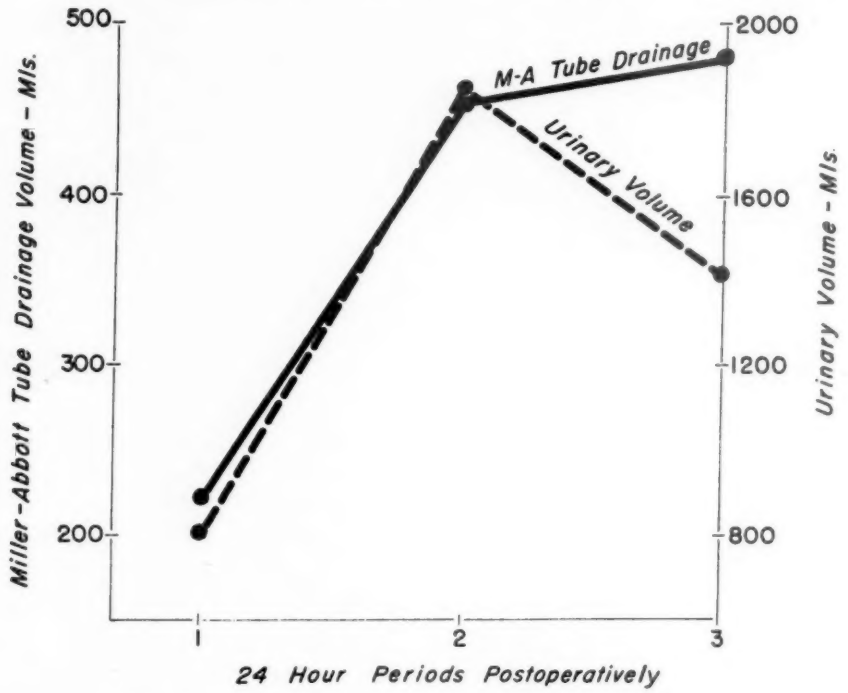

Fig. 1.—Correlation of postoperative urinary volume with volume of intestinal contents aspirated with the Miller-Abbott tube. Note that both are diminished in the early postoperative period. Elsewhere it is shown that during this period adrenocortical activity is increased. (Twenty-nine patients studied.)

the total eosinophil count. This strongly suggests that increased adrenocortical function plays a role in decreasing the volume of intestinal fluid available to aspiration immediately following operation. While the exact mechanism by which this is accomplished is not clear, certain of the adrenocortical hormones are known to effect changes in cell permeability,[1, 3] and it is possible that such action on the cells of the pancreas, liver, and intestinal mucosa in our patients played a part in decreasing the volume of intestinal contents which was observed.

There are other factors which may affect the volume of the various secretions composing the intestinal fluid. Hypotension with inadequate blood flow to the organs concerned could be a factor, but no significant fall in blood

pressure was observed in these patients during operation or in the subsequent period of study. Anoxemia due to poor oxygenation during anesthesia was considered as a possible cause, but the anesthesia charts revealed no evidence that such existed. The effect of the anesthetic mixture per se was probably not a major one, since the results in the patients receiving general anesthesia were not materially different from those having spinal anesthesia. The possibility of significant dehydration in these patients before, during, and after operation would appear to have been eliminated by the fact that each patient was brought to operation in what was considered by the staff to be an optimal state of hydration, the blood loss at operation was replaced as it occurred, and postoperative fluids were administered somewhat in excess of the estimated loss by the several routes.

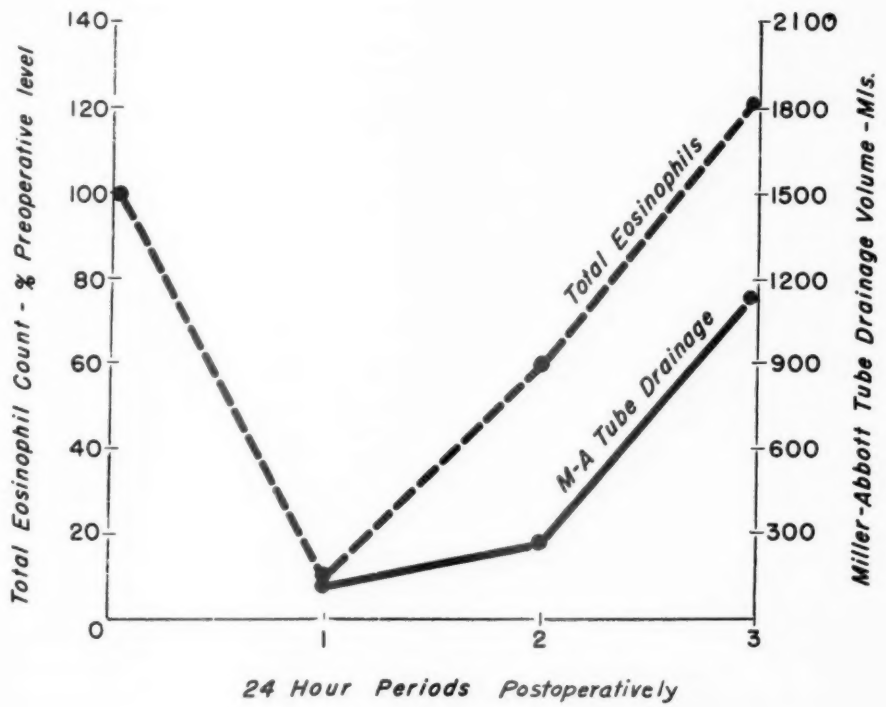

Fig. 2.—Correlation of Miller-Abbott tube drainage with postoperative adrenocortical activity as indicated by the total eosinophil count. Note the fall in the total eosinophil count from the preoperative level of 100 per cent to 10 per cent at the end of 24 hours postoperatively, followed by a gradual return to the preoperative level. During this period of an apparent increase in adrenocortical function, the volume of intestinal secretions aspirated with the Miller-Abbott tube was diminished. (Three patients studied.)

There remains the question of whether or not the moderate manipulation of the small bowel with sponges during exposure of the colon caused localized changes in the bowel wall which resulted in a reduced secretion of succus entericus, and this relationship cannot be entirely excluded. Certainly bowel motility might be altered. Finally, the patency of the Miller-Abbott tube was established before it was passed, and this was assiduously maintained by flushing the tube with 20 ml. of 0.85 per cent sodium chloride solution every 3 hours.

The volume of bile that drained through the T tube in the patients subjected to choledochostomy deserves comment. While it is true that the average volume of bile drained by the 10 patients on the first, second, and third postoperative days was approximately the same, several patients drained little bile during the first 48 hours. This was followed by a profuse flow on the third day. Though no conclusions can be drawn from the data available, the biliary surgeon has long been familiar with the occasional patient whose T tube for no apparent reason fails to drain more than a minimal quantity of bile during the first 24 to 48 hours after exploration of the common bile duct, but then drains freely thereafter. Often no technical imperfection can be found. Could this apparent lag in bile secretion by the liver represent a response of this organ to the hormonal messengers initiated by trauma to the organism?

TABLE I. CUMULATIVE DATA: URINE AND MILLER-ABBOTT TUBE SUCTION VOLUMES DURING THE FIRST THREE 24 HOUR PERIODS POSTOPERATIVELY

	FIRST 24 HOURS		SECOND 24 HOURS		THIRD 24 HOURS	
PATIENTS	URINE VOLUME (C.C.)	M-A TUBE SUCTION (C.C.)	URINE VOLUME (C.C.)	M-A TUBE SUCTION (C.C.)	URINE VOLUME (C.C.)	M-A TUBE SUCTION (C.C.)
G. M.	550	30	900	0	1070	780
D. B.	550	0	735	260	2090	380
M. L.	650	150	2925	65	2190	430
W. H.	1700	0	2650	10	1500	0
M. W.	780	0	1550	300	780	130
L. H.	1200	100	3060	180	1900	280
A. L.	650	290	1910	250	360	60
C. F.	250	400	2100	325	1600	400
W. C.	300		1100	260	1750	380
T. J.	750	250	2400	710	1400	1600
S. K.	1850	540	1770	1200	820	100
M. G.	650	100	1200	175	700	
N. G.	315	265	1860	350	750	650
J. S.	640	1350	1100	2400	1200	
P. A.	1100	50	1750	100	2080	10
H. S.	350	300	1700	480	2550	300
F. P.	1145	375	2320	670		
D. D.	500	330	790	600	1060	210
G. K.	1050	0	2000	760	1200	600
P. D.	700	380	2500	660	1200	800
C. L.	1700	30	1050	295	1300	220
M. R.	200	300	790	800		
S. Z.	900	280	1350	300		380
T. L.			1950	100		
D. L.	155	20	1750	50	900	500
M. G.	950	510	2275	930	920	840
B. B.	1305	50	2520	110	730	1960
E. G.	1150	80	1755	620	3190	1000
M. C.	1180	260	4385	75	2555	400

CLINICAL IMPLICATIONS OF RESULTS

The question of whether or not effective bowel decompression is being maintained postoperatively constantly confronts the abdominal surgeon. The advent of the Miller-Abbott tube, used in conjunction with continuous suction, has materially advanced the margin of safety in small and large bowel surgery. Even in the presence of a competent ileocecal valve, small bowel

decompression is of considerable aid in preventing distention of the colon with attendant strain on the suture line of the anastomosis. The basis for this beneficial effect of small bowel decompression in preventing colon distention in the presence of a competent ileocecal valve is the fact that the major cause of postoperative gastrointestinal distention is swallowed air.[6] This gas is removed by suction before it reaches the cecum to become trapped between the competent ileocecal valve and the anastomosis temporarily occluded by edema.

The surgeon is often dismayed when the tube in the small bowel does not drain what he considers an adequate volume of small bowel contents in the immediate postoperative period. His anxiety may lead him to withdraw a tube which is in actuality accomplishing its purpose. However, our results indicate that once the patency of the tube is established, so long as the abdomen is not distended and the patient remains free from all but moderate incisional pain, the actual volume of fluid aspirated by the tube is relative'y unimportant. If these criteria are fulfilled, then the intestine is in all likelihood decompressed.

CASE REPORTS

The following 2 cases will serve to illustrate the point.

CASE 1.—This 66-year-old white man had a carcinoma of the sigmoid colon resected with primary end-to-end anastomosis under spinal anesthesia. The Miller-Abbott tube in the ileum drained only 100 ml., 180 ml., and 280 ml. during the first, second, and third 24 hour periods postoperatively, respectively. During this period the urinary volume was likewise diminished. The surgeon was alarmed and considered withdrawing the Miller-Abbott tube so that another might be passed. Yet, the patency of the tube had been established repeatedly, the abdomen was soft and flat, and the patient had no pain. Thus the decision to temporize was made. Presently he began to pass gas and feces by rectum and his recovery was uneventful.

CASE 2.—This 52-year-old white man had a carcinoma of the right colon resected with primary ileocolic anastomosis under general anesthesia. The Miller-Abbott tube in the upper ileum drained a net amount of only 30 ml., 5 ml., and 780 ml. during the first, second, and third 24 hour periods postoperatively, respectively. The urinary volumes for these 3 periods were 550 ml., 900 ml., and 1,070 ml. The failure of the tube to drain more than a minimal volume in the first 48 hours following operation was a matter of grave concern to the staff. Nevertheless the patient was comfortable, exhibited no abdominal distention, and had a smooth recovery.

SUMMARY AND CONCLUSIONS

1. Twenty-nine patients subjected to colon resection have been studied with respect to urinary volume and volume of small bowel contents aspirated with the Miller-Abbott tube during the first three 24 hour periods postoperatively. In 3 patients the state of adrenocortical activity following the trauma of anesthesia and operation was estimated by means of the total eosinophil count and these findings were correlated with rates of excretion of urine and intestinal fluid. It was found that when adrenocortical function was maximal, the volume of urine excreted and the volume of intestinal contents aspirated were diminished.

2. The flow of bile may be retarded immediately following anesthesia and operation, but this is not a consistent finding.

3. Regardless of the volume of fluid aspirated by the Miller-Abbott tube in the postoperative period, if the patient remains free of abdominal pain and distention the bowel is probably decompressed.

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SOLITARY DIVERTICULUM OF THE ASCENDING COLON

Case Diagnosed Before Operation

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TO DATE 58 cases of a solitary diverticulum of the cecum or ascending colon have been reported in the English and American literature.[1-5, 7, 8] In nearly all of these the diverticulum was found in the cecum and in all but one or two there was acute or chronic inflammation. Undoubtedly many others have been encountered but not recorded. Solitary diverticula of the large bowel, especially in this region, are uncommon. The importance of this lesion lies in its close clinical resemblance to other far more common diseases of the right colon, such as acute appendicitis, carcinoma, and tuberculosis.[3-5, 8] Thus it usually poses an extremely difficult problem in differential diagnosis. The pathogenesis and pathologic anatomy of colonic diverticula have been thoroughly studied and described by others.[6, 7] Treatment has varied from inversion or excision of the diverticulum to resection of the cecum and ascending colon. Where feasible, the less radical operation is recommended.

The case to be reported is unusual in that it was possible by thorough investigation to establish the diagnosis before operation. Also local constriction of the colon at the site of the diverticulum had resulted in a partial obstruction.

CASE REPORT

Mrs. E. M., a short, obese, 56-year-old woman, was first seen on Dec. 30, 1949, complaining of belching, flatus, sour eructations, and heartburn for 6 months, with vague distress in the right side of the abdomen. The past history was unremarkable except for a chronic bronchitis and the passage of a left ureteral stone 11 years previously, at which time she had suffered an attack similar to the present. Physical examination was negative except for the abdomen, which revealed slight deep tenderness and spasm to the right of the umbilicus but no mass. After a negative intravenous pyelogram, a barium enema (Fig. 1) showed a large diverticulum of the ascending colon some 2 inches proximal to the hepatic flexure. The diverticulum seemed to contain a polypoid tumor or feces and this appearance persisted after evacuation. There was also a persistent constriction of the colonic lumen to one-half or less of its normal diameter opposite the base of the sac. No other diverticula were seen.

After preparation of the patient with Sulfathaladine, operation was performed at the Lawrence Hospital, Bronxville, N. Y., on Jan. 5, 1950. A sea of fat presented itself when the peritoneum was opened. The omentum was a huge apron 2½ inches thick, while the entire ascending and transverse colon was completely invested with fat which filled the leaves of the short mesocolon, effectually obscuring all normal landmarks. The diverticulum was buried in a large mass of indurated fat which was firmly bound to the lateral abdominal wall with dense adhesions. In view of the local chronic inflammation and the known constriction of the bowel lumen at the site of the diverticulum segmental resection was deemed safer than any attempt to invert or excise the sac. Accordingly, the cecum and ascending colon were removed with the diverticulum en bloc and an ileotransverse colostomy was established.

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†Deceased.

*Associate Attending in Medicine.

B.

A.

Fig. 1.—*A*, Preoperative roentgenogram showing large solitary diverticulum of the ascending colon causing constriction of the bowel lumen; *B*, postoperative roentgenogram.

The postoperative course was uneventful save for the appearance of a small fecal fistula which closed spontaneously. Subsequently an abscess of the abdominal wall developed lateral to the operative scar. This subsided promptly upon incision and drainage and the patient has remained well since.

Roentgenograms of the resected specimen filled with barium after operation showed the diverticulum, the thick wall of fat in which it was buried, and the local constriction of the colon (Fig. 2). The gross and microscopic appearance of the specimen is shown in Fig. 3. The pathologic report was as follows: The gross specimen consisted of 18 cm. of terminal ileum, a cecum, and 25 cm. of ascending colon. Fifteen centimeters distal to the colic valve was a diverticulum 21 mm. in length having an ostium 13 mm. in diameter. The lumen of the diverticulum was wedge-shaped, the ostium representing the base. The internal surface was smooth; a fat pad surrounded the distal tip. Histologic study showed that the muscularis gradually thinned at the neck of the diverticulum and at the periphery the wall consisted merely of mucosa and serosal fat. A clump of lymph follicles in the serosal fat showed fibrotic changes.

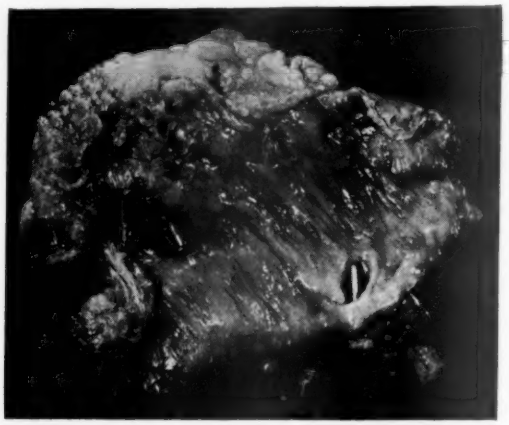
Fig. 2.—Roentgenogram of resected specimen filled with barium after operation. Note the thick wall of fat and the local constriction of the colon.

SUMMARY AND CONCLUSIONS

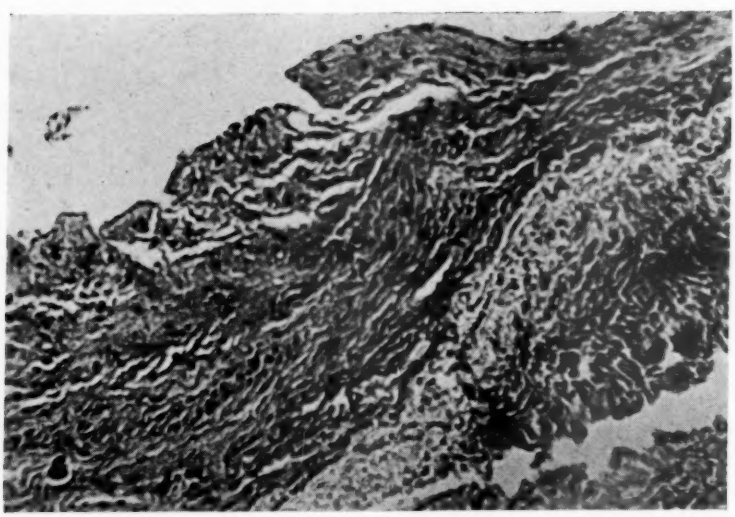
1. A case of solitary diverticulum of the ascending colon which was diagnosed before operation is reported and the literature is briefly reviewed.

2. Although the occurrence of this lesion is uncommon it should be considered in the differential diagnosis of acute appendicitis, regional ileitis, and carcinoma or tuberculosis of the cecum. Accurate preoperative diagnosis is usually impossible.

3. Inversion or excision of the diverticulum is ordinarily sufficient but segmented resection of the colon may at times be indicated.



A.



B.

Fig. 3.—*A*, Photograph of gross specimen, opened, showing mouth of diverticulum; *B*, photomicrograph of section through the junction of diverticulum and normal colon. Note loss of muscle coat in the diverticular wall.

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SPONTANEOUS RUPTURE OF THE ESOPHAGUS

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BY THE term "spontaneous rupture of the esophagus" is meant the rupture of an esophagus presumed to have been previously normal. Spontaneous rupture of the esophagus is considered a rare event and recovery from such a catastrophe is even more rare. The first description of the lesion was made by Boerhoave in 1723, as reported by Lynch,[1] and the first diagnosis during life was made by Walker[2] in 1914. At the present time the diagnosis has been made many times before death but seldom early enough to achieve any success with treatment. It was not until 1947 that the first case of recovery was recorded in the literature. Barrett[3] reported a case in which the tear was closed with a purse-string suture and the patient recovered. At about the same time Olsen and Clagett[4] reported a case in which a patient recovered following closure of the tear. The case we are presenting is of interest because it is one of the first cases reported in which closed thoracotomy and drainage, in conjunction with the newer antibiotics, were instituted early in the course of the disease and the patient recovered.

REPORT OF A CASE

F. C., a 34-year-old Negro man, was admitted to the John Gaston Hospital on March 19, 1950, complaining of severe pain in the abdomen and chest. He stated he had always consumed a large quantity of alcohol and had been drinking heavily the previous night. On the day of admission to the hospital the patient felt well on awakening except for a headache. He ate a good breakfast and at 10:00 A.M. suddenly became nauseated and began to retch violently and vomit. The vomiting subsided but recurred again a few minutes later. During this episode of vomiting he was seized with severe, excruciating pain across the lower and upper abdomen which radiated up into the left chest and left flank. The pain was constant and was more severe on swallowing. The patient's condition gradually became worse throughout the day and when he was seen in the emergency room at 8:00 P.M., 10 hours after the onset of the pain, he appeared acutely ill, dehydrated, and in a moderate degree of shock. Physical examination revealed a well-developed, well-nourished Negro complaining of severe pain across the chest and abdomen. Respirations were 80 per minute and of a grunting, catchy nature, so shallow they were hardly perceptible. A small amount of air could be palpated in the subcutaneous tissue in the left supraclavicular region. Auscultation of the chest was negative except for a few râles heard in the left base. Tenderness to pressure was present over the left lower chest. The entire abdomen was rigid, resembling the "boardlike" abdomen of a ruptured peptic ulcer. Tenderness and rebound tenderness were present over the entire abdomen but most marked over the left upper quadrant. Peristalsis was present but hypoactive.

Roentgenogram of the chest was reported as negative except for subcutaneous air noted in the left side of the neck. A scout film of the abdomen was negative. A diagnosis of spontaneous rupture of the esophagus was made. Another chest film was made after the patient had been given 2 ounces of Iodochlorol to swallow. The diagnosis was confirmed when a small amount of the radiopaque oil escaped into the lower mediastinum and left pleural space.

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On admission to the ward there was very little change in his condition except that the subcutaneous emphysema previously noted could not be demonstrated again. A left thoracentesis was performed and several ounces of a chocolate-colored fluid containing small particles of grayish-white material were obtained. There was no odor to the fluid and it was found to be alkaline in reaction. The patient was taken immediately to the operating room and under local anesthesia a rib resection thoracotomy was done. Approximately 500 c.c. of a brownish, odorless fluid containing numerous small particles of solid material were removed from the pleural cavity. A large rubber tube was inserted and connected to an underwater drainage bottle. The patient was given 500 c.c. of blood on the operating table and a transfusion every other day for a week.

After returning from the operating room a Levine tube was passed into the stomach, its position confirmed by roentgenogram, and gastric suction was begun. The patient was given 3,000 c.c. of 5 per cent glucose solution containing 8.5 Gm. of sodium chloride intravenously each day. Nasal oxygen was started and the patient was given nothing by mouth. He was given 300,000 units of penicillin every 3 hours, ½ Gm. streptomycin every 4 hours, and 2 Gm. sodium sulfadiazine intravenously 3 times daily. On the first postoperative day gastric suction was discontinued but the Levine tube was left in the stomach. The patient was given 1 Gm. of aureomycin in an ounce of water by mouth, which was continued every 4 hours. He experienced pain in the chest each time he swallowed this medication. The patient was acutely ill with rapid, grunting, shallow respirations, a fever of 103° F., and a pulse rate of 140. On the second postoperative day 600 c.c. of fluid were aspirated from the right chest. The fluid was similar to that removed from the left chest during the operation and *Streptococcus viridans* was cultured from both fluids. No further aspirations of the right chest were necessary. On the fourth postoperative day the patient appeared improved, complained of little pain in the chest, and the tenderness and rigidity of the abdomen were less marked. He was then given methylene blue by mouth, none of which appeared in the drainage from the thoracotomy tube. Water and a 3,000 calorie liquid formula were given through the Levine tube daily. The patient continued to improve but remained critically ill for several days. By the eleventh postoperative day the temperature was 99° F. and the pulse was normal. The patient felt well and drainage from the thoracotomy tube had ceased. The Levine tube was removed and water was given by mouth, and on the fourteenth postoperative day a soft diet was given. The patient was discharged at the end of 5 weeks. When last seen 5 months after the illness the patient appeared to be in good health and stated that he had "quit drinking and was going to church on Sunday."

DISCUSSION

In most cases of spontaneous rupture of the esophagus recorded in the literature no previous history is obtained to lead one to suspect disease of the esophagus. There is a striking similarity, however, in the history, physical findings, and clinical course of these patients. Most of the patients are men between the ages of 35 and 45 who experience severe abdominal and chest pain during episodes of vomiting following an alcoholic debauch. The pain is constant and radiates across the upper abdomen and lower chest and into the back. Some degree of shock is usually present. Respirations are short, rapid, and shallow, and there is a splinting of both the chest and abdomen with resulting dyspnea and even cyanosis in some instances. Swallowing either liquids or solid food causes severe pain in the chest. Tenderness and rigidity are usually present across the upper abdomen and the chest is clear for a short period of time after the rupture. These signs usually continue for the first few hours and then there is a rupture into the pleural cavity which

usually occurs on the left side. Only 2 cases were found in the literature in which the patient was operated upon before rupture occurred into the pleural cavity.[5, 6] The physical signs at this time are usually those of pleural effusion on the left side. After the first few hours subcutaneous air may be palpable in the neck or noted on roentgen examination. This finding has been present in over 80 per cent of the cases reported but it may be transient and can be easily overlooked. Also at this time there is some rigidity of the abdomen, described as "boardlike" in many instances. An exploratory laparotomy with a preoperative diagnosis of perforated peptic ulcer has been performed in about 15 cases due to this finding.

When a patient gives a history of sudden pain in the chest, especially the lower chest or upper abdomen, which has occurred during vomiting, spontaneous rupture of the esophagus should be considered. Although there are no early symptoms or physical findings characteristic of spontaneous rupture of the esophagus, the presence of air in the neck of a patient with this history is diagnostic. With the exception of trauma no other condition except possibly primary spontaneous pneumothorax will produce mediastinal emphysema and it is rare in this condition.

Usually the diagnosis may be confirmed by giving a radiopaque oil by mouth and demonstration of its presence in the mediastinum by roentgenogram. Barium should not be given if a perforation of the esophagus is suspected. The fluid obtained from the pleural cavity usually produces an acid reaction, our case being the only one reported in which an alkaline reaction was obtained.

The condition of those patients who are not treated surgically deteriorates progressively and the majority will die within 2 days after the rupture occurs. There have been 2 cases reported in the literature where a localized empyema and mediastinal abscess developed, the perforation closed spontaneously, and recovery followed drainage of the empyema or mediastinal abscess.[7] Most surgeons agree that the most important single factor in the treatment of spontaneous rupture of the esophagus is the time interval between rupture and surgery, but not all agree as to the most effective type of operative procedure in these cases. Generally, 2 types of operative treatment are advocated: thoracotomy with drainage, and thoracotomy with repair of the laceration.

Of the 9 patients reported recovering following surgery, 3 were treated with thoracotomy and drainage, 5 by primary repair of the laceration, and 1 by thoracotomy and drainage, the laceration being repaired at a later date. Frink[8] reported a case in which repeated thoracenteses were performed and a rib resection with drainage was done 3 weeks after the rupture occurred and the patient survived. One of the 3 patients reported by Lynch[1] was treated by temporary drainage with an intercostal tube followed later by open drainage of the mediastinal abscess and empyema. The perforation was 60 hours old before the diagnosis was made and the patient was acutely ill for 3 months before he recovered. The third case in which recovery occurred following

drainage was reported by Gardner.[5] The diagnosis was made before rupture into the pleural cavity occurred and a posterior mediastinotomy was performed. The patient made a prompt recovery.

Until a relatively recent date most patients with spontaneous rupture of the esophagus were treated by thoracotomy and drainage with a high mortality rate. Attempts at closure of the perforation were made by Collis and associates[9] and by Foggitt[6] but the patients died a few hours after the operations were completed. Kinsella's[10] patient was well on the road to recovery only to die of a pulmonary embolism on the ninth postoperative day. It was not until 1946 that Barrett[3] successfully repaired the defect and the patient survived. The next year Olsen and Clagett[4] reported a case in which they successfully closed the perforation only 3 hours after the rupture occurred. Moore and Murphy[11] recently reported an unusual case that was treated at first conservatively with a thoracotomy and drainage and was repaired successfully 3 weeks later. These authors suggested drainage and jejunostomy as the treatment of choice, believing that the perforation will heal spontaneously in most cases. In the cases where healing does not occur operative repair should be undertaken. Scholefield[12] recently reported a case in which repair of the perforation was successful in a patient in whom perforation occurred into both pleural cavities. Two of the 3 cases reported by Lynch[1] were treated successfully by repairing the perforation. He believed that adequate drainage of the mediastinum with closure of the perforation is necessary to cure these patients.

COMMENT

Ideally, the correction of this condition should follow the same lines as the treatment of a perforated peptic ulcer, that is, closure of the defect; however, the decision to undertake such an operation may not always be good judgment. The patient is acutely ill, in shock with a reduced vital capacity, and is a poor candidate for anesthesia or surgery even though some improvement may occur when the pleural cavity is opened and compression of the mediastinal structures is released. It has been emphasized that the time interval is an important factor. When the patient is seen only a few hours after the perforation and the diagnosis is made early, the tear should be closed. This is best performed through a transthoracic approach under an endotracheal anesthetic. In only 1 case was the repair attempted through the abdomen and this was unsuccessful.[6] In the patient in whom the diagnosis is made later and the patient is critically ill, a rib resection and drainage of the pleural cavity with the concomitant use of the newer antibiotics offers a better chance of recovery than was formerly the case. Gastric suction with a Levine tube in the stomach should be continued for several days. It is important to make a scout film of the abdomen after the Levine tube is inserted, one case having been reported in which the tube was placed in the mediastinum.[13] When the suction has been discontinued the patient may be fed through the

Levine tube and a gastrostomy or jejunostomy is not necessary. The other side of the chest should be watched carefully and thoracentesis should be performed if fluid develops, and the pleural cavity drained if necessary.

SUMMARY

1. The clinical picture of spontaneous rupture of the esophagus is presented.

2. A case of spontaneous rupture of the esophagus with recovery is presented in which the treatment consisted of thoracotomy with drainage with intensive antibiotic therapy and chemotherapy.

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THE MANAGEMENT OF FAMILIAL POLYPOSIS OF THE COLON

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IN RECENT years there has been an increasing number of cases of familial polyposis of the colon reported. This interesting disease is characterized by the presence of multiple polyps in the large bowel as an inherited Mendelian dominant characteristic.[7] In given instances of the disease the polyps may be few in number or they may be present in thousands; the polyps may be present in isolated regions of the large bowel or they may involve the large intestine diffusely from the ileocecal valve to the anus. The polyps, which may appear at any period of life from childhood to adulthood, tend to progress from benign adenomatous tumors into carcinoma. Therefore, the unfortunate individual in a family afflicted with this disease lives with the continued possibility of developing this disease, and, once having developed it, of dying of carcinoma of the large bowel unless adequate surgical treatment is administered.

There are 2 customary methods of surgical management of familial polyposis in which a carcinoma has not developed in the rectum. The more conservative treatment is resection of the colon with anastomosis of the terminal ileum to the lower large bowel, thereby retaining a variable amount of the rectum and an unimpaired anal sphincter. The other form of treatment is removal of all the large bowel, colectomy plus abdominoperineal excision of the rectum.

The argument in favor of retaining a rectal segment should be advanced only after certain criteria have been considered. Various of these criteria have been mentioned by others[1, 5] as factors influencing the choice of procedure. I would like to propose four criteria as the determining factors in the choice of operation to be employed. These criteria are: (1) there must be no carcinoma present in the rectum at the time of inception of treatment; (2) the rectal segment remaining after surgery must be amenable to complete visualization with a proctoscope; (3) this segment must be completely cleared of polyps; and (4) repeated examinations of the rectal segment with biopsy and destruction of new polyps (if any) must be performed every 3 months for the remainder of the patient's life.

When these criteria are met so as to allow retention of the rectum, the advantage of this procedure is that the patients lead a relatively normal existence. The routine follow-up proctoscopic examinations are a minimal inconvenience as compared with the daily ministrations to an ileostomy, even though the skin-covered protruding ileostomy stoma described by Dragstedt[2] is a great improvement over the old flat stoma. This is a point of added importance when one reflects that patients with this disease frequently are candidates for surgery in the first four decades of life, the period during which marriage and domestic and social activities are of considerable importance. However, the possibility of developing carcinoma in the rectal stump remains as a disadvantage.

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When the treatment consists of colectomy plus proctectomy, the threat of carcinoma of the large bowel has been eliminated at the expense of the care of a permanent ileostomy.

There is considerable conflict of opinion as to which operation is the procedure of choice. There can be no doubt that ileorectostomy with retention of unimpaired sphincter function is more compatible with a normal existence than is total excision of the large bowel with an abdominal ileostomy. The crux of the controversy rests on the questions of (1) whether the four criteria can be met to allow ileorectostomy, and (2) how great is the potential danger of carcinoma following ileorectostomy once the four criteria have been satisfied. Particularly might one be loath to perform proctectomy in each case when a patient who underwent colectomy with ileosigmoidostomy for polyposis of the colon in 1900[8] was reported living and well 19 years later without apparent follow-up examinations in the interim. Likewise, a patient who underwent a similar operation for a like condition in 1914[15] was reported living and well in 1936.*[9] Obviously, the reports of single cases carry little proof; the preferred method of assaying these questions would be a detailed statistical study of all the known cases of familial polyposis which would include adequate follow-up data. With relatively small individual series of cases reported by different authors over a period of years, such an endeavor would seem to be very difficult. And so I found it when I attempted such a survey, which was productive only in so far as it brought to light approximately 20 unreported cases of this disease.

A questionnaire was sent to surgeons who have had experience with the treatment of familial polyposis. This questionnaire polled their individual opinions as to the proper operative management of familial polyposis. Seventeen surgeons[16] responded with either the answered form or with a courteous letter explaining their ideas on this subject. Three plans of operative management were presented and the surgeon indicated his preference. The questions and responses were as follows:

PLAN OF MANAGEMENT FOR FAMILIAL POLYPOSIS	NUMBER OF SURGEONS PREFERRING PLAN
1. Every case should have a total colectomy, proctectomy, and ileostomy	2
2. Every case should have colectomy with anastomosis of the terminal ileum to the lower large bowel provided that:	
(a) the remaining large bowel contains no carcinoma	
(b) the remaining large bowel is accessible to a proctoscope	
(c) the patient agrees to cooperate for continued follow-up proctoscopic exams with fulguration and excision of polyps	
(d) subsequent proctectomy to be performed if a carcinoma develops in the remaining large bowel	3
3. A compromise between Plans 1 and 2, accepting Plan 1 in cases with extensive polyposis of the rectum and Plan 2 in cases with a mild degree of polyposis of the rectum	12

Truly there is not unaniminity of opinion in this poll, yet this poll does reflect the present-day opinions regarding management of this disease.

*Family history not mentioned in the reports of these 2 cases.

"FAILURES" OF TREATMENT WITH RETENTION OF RECTUM

As the advocates of total excision of the large bowel always point to the reported cases which developed malignant disease in the retained rectal segments, one method of appraising the value of the 4 criteria as a determining factor in the choice of operation would be to scrutinize these "failures." Those cases reported in the literature which developed a carcinoma of the rectum following anastomosis of terminal ileum to lower large bowel have been analyzed in respect to the 4 criteria just presented; this analysis is not a critique of the individual results obtained, as the surgeons in some cases were obviously limited in their treatment by the patients' desires. The appearance of carcinoma in the retained rectal segment is certainly an undesired development in any individual case; however, such a development will not be considered as a failure of the form of treatment unless the 4 criteria had been satisfactorily met. A brief review of such cases follows:

CASE 1 (Reported by Jones[6]).—A 32-year-old woman with extensive polyposis refused ileosigmoidostomy and colectomy until 1931, although she had experienced bloody diarrhea since 1923. During that period she had had over 400 polyps destroyed from below and a cecostomy performed for obstruction from edematous polyps. Although malignant disease was not demonstrable in the myriads of polyps in the resected specimen, the patient died in 1935 with a metastatic tumor mass in the retroperitoneal nodes. Postoperative proctoscopic examinations were performed and apparently there was never any suspicion of carcinoma in the rectum.

CASE 2 (Reported by Pfeiffer and Patterson[12] [Case 4]).—A 42-year-old man with diffuse polyposis of the colon (family history negative for intestinal complaints) underwent resection of an obstructing carcinoma of the sigmoid in 1943. The polyps in the lower segment were subsequently fulgurated and colectomy with ileorectosigmoidostomy performed. A large retroperitoneal mass was palpable on rectal examination 17 months following his original resection. Examinations of the lower segment did not reveal carcinoma before or after the resection.

Comment.—From the available data relative to Cases 1 and 2, it seems that one should assume the metastatic retroperitoneal masses to have been secondary to primary carcinomas in the resected colons, not to malignant degeneration in the rectal segments. This was Jones' opinion relative to his case in which he endorsed the procedure employed but believed earlier surgery had been indicated. Pfeiffer[11] had a similar impression regarding Case 2. Neither of these cases can be termed a failure by virtue of retention of the rectum inasmuch as all evidence points to the fact that there was no carcinoma of the rectum.

CASE 3 (Reported by McKenney[10] [Patient F. S.]).—A 35-year-old man who had been treated for polyposis by ileorectosigmoidostomy and colectomy died of carcinoma of the rectum. The primary tumor developed in an area of extensive polyposis which was above the area from which 600 polyps had been cleared 4 years earlier. Interim follow-up was not mentioned.

Comment.—The photograph of the autopsy specimen in the original article clearly demonstrates the primary tumor above the area of clearing. The case should be considered as incompletely treated, not as a failure of the mode of management.

CASE 4 (Reported by Ravitch and Sabiston[14]).—A 16-year-old patient with diffuse polyposis (familial history not given) and extensive rectal involvement underwent rectal clearing, colectomy, and ileosigmoidostomy in 1937. Proctoscopy was performed in 1938; rectal examinations were performed on subsequent hospitalizations for a genito urinary complaint. In 1945, the patient re-entered the hospital with an obstructing carcinoma of the rectum. The rectum was resected but the patient died of carcinomatosis 8 months later.

Comment.—From the facts presented, one cannot say that this case is a failure of the method employed as it is certain that only one proctoscopic examination was performed between 1937 and 1945. To conform with the criteria presented, a minimum of one proctoscopic examination should have been made every 3 months for adequate control of polyps in the rectal segment.

CASE 5 (Reported by Boehme[1] [Case 1]).—A 48-year-old woman underwent colectomy and ileorectosigmoidostomy in March, 1947. The retained rectal segment contained dozens of isolated polyps plus 2 larger matted sessile polyps. Subsequent repeat biopsy of the matted polyps revealed malignant change and proctectomy was performed.

CASE 6 (Reported by Guptill[3]).—A 19-year-old boy was originally seen in 1936 for extensive polyposis, the rectum being nearly filled with polyps. By 1939, hundreds of polyps had been fulgurated, and colectomy and ileorectostomy had been performed. In 1939, a biopsy reported malignant changes and fulgurations were continued. In August 1941, scarring from the extensive fulgurations necessitated rectal dilatation. The following month a biopsy showed definite carcinoma of the rectum. The rectum was resected but the tumor was then incurable.

Comment.—Cases 5 and 6 share a common problem of analysis. In reporting Case 5, Boehme stated that his original biopsy of the matted polyp was not deep enough. In Case 6, the patient's rectum is described as being nearly filled with polyps and subsequently hundreds of polyps were destroyed by fulguration. With extensive involvement, diffuse or in the form of isolated matted polyps, it is extremely difficult to be certain that carcinoma is not present. It would take an infinite number of biopsies to prove that fact, realizing that malignant changes may appear in any part of the adenoma[4]; likewise it demands an extraordinary number of procedures with the passage of considerable time to destroy such tumors to the point where one could claim complete clearing of the rectum. Rectal stenosis in Case 5 attests to the degree of fulguration required. Neither the first nor the third criterion can be satisfied in these cases and these cases are therefore not failures from the standpoint of this analysis.

REPORT OF CASES*

CASE 1 (Barnes Hospital, St. Louis, Mo., No. 104595).—L. R., a 48-year-old white woman, underwent a successful obstructive resection of an obstructing carcinoma of the left colon in 1937 (operation performed elsewhere). In 1943 an occasionally functioning fecal fistula at the site of the resection was closed at Barnes Hospital with no other abnormalities noted. In 1947 the patient noted an 11 pound weight loss plus occasional flecks of dark blood in the stools. Proctoscopic examination revealed several small, inconspicuous rectal polyps; barium enema disclosed a tumor of the right colon. At that time, a family history was first elicited to the degree that the patient's mother, one sister,

*The first 2 patients were treated by the author as Resident Surgeon at Barnes Hospital.

and one brother had died of carcinoma of the large bowel before the age of 50. In June, 1947, a right colectomy was performed when exploration revealed no tumor other than a primary carcinoma of the right colon. Pathologic examination showed a carcinoma of the right colon, metastasis to regional nodes, and diffuse polyposis of the colon included in the specimen. After a 17 month observation period in which no evidence of metastatic disease or new tumor could be ascertained, the remaining colon was excised with anastomosis of the terminal ileum to the high rectum. No evidence of carcinoma was found at time of laparotomy or in the surgical specimen, which contained multiple polyps. During the 18 months since her last resection, this patient has had 6 proctoscopic examinations; a total of 5 sessile polyps 3 mm. in diameter has been fulgurated.

CASE 2 (Barnes Hospital, St. Louis, Mo., No. 108909).—M. J., a 26-year-old white woman, who is a niece of the patient in Case 1, had experienced intermittent bouts of intestinal cramping, diarrhea, and some bright blood on stools for 6 years. Five years previously, a diagnosis of amoebic dysentery had been made; however, specific amoebicidal medications had produced no therapeutic effect. At time of hospitalization, 5 small sessile rectal polyps were visualized at proctoscopy; a barium enema outlined a relatively large polyp in the sigmoid and another in the splenic flexure of the colon. On July 9, 1948, a laparotomy was performed; inspection of the mucosa of the colon through a small incision revealed diffuse small, sessile polyps whose presence had been debatable on the x-ray film. With the diagnosis of familial polyposis confirmed, a right colectomy was performed with anastomosis of the terminal ileum to high rectum. Three weeks later, the remaining colon was removed and the rectosigmoid turned in above the anastomosis. No evidence of carcinoma was found at either operation or on examination of the surgical specimen. During the 24 months following surgery this patient has undergone 11 proctoscopic examinations at the clinic. Twenty-six polyps have been destroyed during this period; one polyp was a small pedunculated polyp, while the remaining 25 polyps were 2 to 3 mm. mucosal protrusions.

CASE 3 (St. Joseph's Hospital, Alton, Ill., No. 50-1281).—H. V., a 25-year-old farmer's wife, mother of 2 children, is the niece of Patient 1 and a cousin of Patient 2. She had experienced intermittent mild diarrhea, sometimes accompanied by blood, for about 5 years. Proctoscopic examination revealed 25 small polyps in the rectum; barium enema demonstrated changes consistent with the diagnosis of polyposis. On April 14, 1950, a total colectomy was performed and the terminal ileum anastomosed to the high rectum. The patient was discharged from the hospital on the tenth postoperative day. On May 29, 1950, she returned to the hospital for biopsy and fulguration of the polyps in the rectal segment; she is to return again in 3 months. A combination suction-fulguration tip as described by Dr. Boehme was used. This instrument made the destruction of the polyps unusually simple, safe, and complete. No evidence of carcinoma was found in either the surgical specimen or the biopsy specimens.

Comment.—Each of these 3 cases has met the 4 criteria which I feel are important prior to colectomy with retention of a rectal segment. In each case there has been (1) no malignancy present in the rectum prior to treatment. (2) the rectal segment remaining after surgery can be visualized in entirety through a proctoscope, (3) the remaining polyps have been destroyed, and (4) regular follow-up examinations are being maintained. These patients have had 2 bowel movements daily; their disease has placed no burden upon their lives other than regular follow-up examinations. The first 2 patients have been married subsequent to their operations; the last patient has continued the active life of a mother and farm wife.

DISCUSSION

When one attempts comparison of the results of the 2 usual forms of operative management of familial polyposis, reliance is generally placed on previously published cases. It should be remembered that a case is successful only if the patient, living and well after a variable period of time subsequent to colectomy with retention of some part of the rectum, continues to be free of carcinoma of the rectum in the future. Also, from the standpoint of this study, similar treatment of a patient who subsequently developed carcinoma of the rectum cannot be classified as a failure of the form of management unless the previously mentioned 4 criteria have been met. Six possible failures of the conservative type of treatment are present in the literature. Scrutiny of these reveals none of the cases to have met the 4 criteria and therefore none may be considered to represent failures of the conservative form of treatment if the 4 criteria are considered as an integral part of the management. Patients who have extensive involvement of the rectum are not candidates for operation with retention of a rectal segment if satisfaction of the 4 criteria be considered necessary.

If one accepts the use of the 4 proposed criteria as the determining factor as to the type of operation to be employed, there are still a number of decisions which must be made by the individual surgeon. Exactly when the degree of rectal involvement does or does not allow satisfaction of the criteria for the conservative operation is up to the surgeon's judgment. Matted polyps or extensive isolated polyps should not allow retention of the rectum. The amount of rectum allowed to remain may vary with individual surgeons and individual cases. One surgeon who is listed as preferring the second treatment plan in the questionnaire survey invariably leaves only 2 inches of a rectal segment; other surgeons leave the entire rectum. Surgeons have also presented arguments favoring the suitability of an end-to-end anastomoses. With the passage of a few months, the small blind pouch of rectum above a side-to-side anastomosis shrinks down while the other elements dilate; the result is a conformity similar to an end-to-end anastomosis without blind spots for proctoscopic visualization. However, mucosa should not be buried in the closure of the rectal stump if a side-to-side anastomosis is used. I believe the type and level of anastomosis employed should be chosen by the surgeon from the standpoint of technical facility and existing pathology in the individual case.

It is fully realized that the mode of management of familial polyposis of the colon may cease to be a problem if the operation devised by Ravitch and Sabiston[14] gives satisfying results. An anal ileostomy with preservation of sphincter function would combine the advantages of the 2 customary procedures with elimination of the disadvantages. At present, this procedure has been performed 5 times for familial polyposis of the colon,[13] and time may solve the present controversy by routinely replacing the customary procedures with anal ileostomy. However, the test of time may also disclose some disadvantages incident to the procedure.

The opinions and material sought from other surgeons by questionnaires were limited to cases with positive family histories. Diffuse polyposis is probably

the same disease in the form of a mutant without a positive family history. However, information regarding diffuse polyposis was intentionally not included in the questionnaires because of the possibility of accumulating opinions regarding the postinflammatory type of multiple polyposis or the infantile type of multiple polyps, both of which must be considered as different diseases. Cases of diffuse polyposis are included in certain references to the literature; particularly in appraising the "failures" of the conservative type of treatment did it seem advisable to include all possible instances, utilizing cases with and without a positive family history. All references and cases refer to familial polyposis unless the lack of a positive family history has been specifically noted. Impressions presented in this paper should be applicable to diffuse polyposis as well as familial polyposis.

SUMMARY

1. The 2 common methods of management of familial polyposis of the colon have been compared.

2. A strict set of 4 criteria has been enumerated which should be satisfied prior to the adoption of colectomy with retention of the rectum.

3. A poll of 17 surgeons experienced in the treatment of this disease revealed that the majority of the surgeons (70 per cent) prefer management by means of colectomy with anastomosis of the terminal ileum to the lower large bowel if the preceding criteria are met. A minority of surgeons would perform a proctectomy in each case; another minority of surgeons would not perform proctectomy until carcinoma of the rectum is histologically demonstrable.

4. Analysis of 6 possible failures of the conservative manner of management reveals no true failures of the method if satisfaction of the 4 criteria is considered an integral part of that form of management.

5. It is particularly the patient with extensive involvement of the rectum in whom the 4 criteria cannot be met for adoption of the conservative method.

6. The apparent advantages of an anal ileostomy with preservation of sphincter function have been mentioned. Continued study of this operation will divulge if disadvantages may be incident to this procedure.

7. The impressions presented regarding familial polyposis are applicable to diffuse polyposis of the colon, but not to other forms of polyps of the colon.

8. Three cases of familial polypsis of the colon treated by colectomy with retention of the rectum are presented.

Appreciation is expressed to those surgeons whose personal communications form an interesting and valuable part of this paper.

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HERNIATIONS OF THE CERVICAL INTERVERTEBRAL DISC AND SCALENOTOMY

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THE clinical and pathologic entity of herniation of the cervical intervertebral disc has been clarified within the past decade. From the beginning, much attention has been paid to the differential diagnosis between lateral herniations of the lower cervical intervertebral discs and the scalenus anticus syndrome. The general opinion is that these are distinct entities and, with careful study, the differentiation can be made with certainty in the majority of cases. A few observers have expressed the opinion that the radiating brachial plexus pain observed in hernations of cervical intervertebral discs may in part be due to spasm of the scalenus anticus muscle, possibly secondary to the root irritation. We wish to report a series of cases supporting this concept that the scalenus anticus and cervical disc syndromes are not necessarily mutually exclusive and comment on the implications concerning therapy for patients with upper extremity pain.

CASE 1.—S. Z., a 50-year-old bookkeeper and housewife, had abrupt onset of pain in the right upper extremity while sitting at her desk in January, 1947. It persisted in a mild form until June, 1947, when it became definitely worse. By August, 1947, the pain was so severe that she was unable to use the extremity. However, it improved thereafter, and for three or four weeks preceding Thanksgiving, 1947, she was relatively free of pain. On Thanksgiving Day she picked up a 20 pound turkey, following which she had a recurrence of pain which persisted until she was examined in January, 1948. At this time the pain was constant, but not as severe as it had been the preceding summer. The pain was in the nature of a severe ache, but with definite paroxysms of increased pain, starting in the lateral aspects of the right arm, radiating to the lateral part of the elbow, then to the dorsum of the forearm and into the right thumb and first two fingers. There were also radiations of pain into the right side of the neck and to the area just lateral to the upper six or eight thoracic vertebrae. The pain definitely awakened her from her sleep, and was so annoying that she was willing to contemplate a serious operation for its relief.

She recalled a fall three years before, having slipped on the ice and fallen backward, striking the back of the head, shoulders, and lower part of the spine. She had some pain in the neck, shoulders, and spine for several days, but was subsequently free of pain until her present illness.

The patient had hypertension for several years and there was definite evidence of cardiac enlargement and myocardial damage.

Examination revealed a short, obese woman in obvious discomfort. There was possibly less power in the right hand grip than the left and the right triceps reflex was absent. No clear sensory change was noted. She was definitely tender over the right scalene muscle. Pulses were all present and there was no evidence of vascular involvement by the scalene muscle on testing with the several recommended maneuvers. There was generally some restriction of movements of the neck, but the pain was not aggravated by movements of the neck.

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Roentgenogram of the cervical spine revealed a loss of the normal lordotic curve with narrowing of the interspace between the fifth and sixth cervical vertebrae.

Cervical traction was recommended and carried out for three weeks without benefit. A major procedure such as a cervical laminectomy seemed hazardous in this obese patient with hypertensive cardiac disease, who was only partially disabled by her pain. Attracted by the tenderness over the scalene muscle, sectioning of this muscle was recommended on the basis of its being a relatively benign procedure.

Early in February, 1948, a right anterior scalenotomy was carried out.

She was immediately relieved of all upper extremity pain, although she had occasional twinges of pain in the right shoulder and neck. She has been seen or heard from at regular intervals since and now, more than two years postoperatively, she is completely content, being entirely free of pain in the right upper extremity and apparently undisturbed by occasional pain in the right shoulder and neck.

CASE 2.—B. M., a 32-year-old veteran, was admitted to the Wilmington Veterans Hospital on Dec. 13, 1948. He complained of pain in the right shoulder, down the arm and forearm, and into the index finger. There was occasional pain in the posterior cervical region. He recalled the onset of this pain in 1944, but that episode lasted only for a short while. He had a recurrence of pain in the summer of 1945; the symptoms continued intermittently, and he was hospitalized on several occasions at various veterans hospitals.

Examination revealed a well-developed young man who held his head slightly to the right. Hand grip on the right was possibly weaker than on the left. There was some hypalgesia of the right index finger. All reflexes were intact and no evidence of vascular involvement of the extremity could be detected.

Roentgenogram of the cervical vertebrae revealed loss of the normal lordotic curve and narrowing of the interspace between the fifth and sixth thoracic vertebrae. There were no cervical ribs, but localized osteoarthritis was present in the region of the fifth and sixth cervical vertebrae.

Cervical traction was applied for a period of about two weeks. He had relief from cervical pain, but continued to complain of pain in the right shoulder and upper extremity. He did not feel the pain was severe enough to warrant a major surgical procedure, but did desire more relief than traction had given him. On Jan. 30, 1949, a right anterior scalenotomy was done after explaining to the patient the possibilities of failure of this procedure and, on the other hand, the relative difficulties of scalenotomy and laminectomy.

He was immediately improved, and returned to work on Feb. 9, 1949, as a salesman. He was last heard from in January, 1950, when he reported freedom from pain in the right shoulder and upper extremity.

CASE 3.—J. W., a 50-year-old man, entered the hospital June 15, 1948, complaining of attacks of painful numbness of the right forearm and entire hand lasting three to four minutes and occurring numerous times daily over a period of four months. He could not recall any circumstance that caused an attack of pain, but raising his hand above his head definitely relieved him during an attack. The pain was often severe enough for him to lose time from his occupation of movie projectionist. Cervical traction prior to admission had been ineffective.

Six years previously he had had pain in a similar distribution over a period of three to four months which responded to chiropractic treatment. There was no history of trauma.

Neurological examination was entirely normal except for a slightly diminished right triceps reflex and the ability to reproduce the pain by pressure over the right scalenus anticus muscles. There were no evidences of vascular involvement in the various tests recommended to increase the pressure of the scalene muscle on the subclavian vessels.

Plain x-ray films of the cervical spine revealed hypertrophic degenerative changes, with slight narrowing of the interspaces between the fifth and sixth and sixth and seventh

cervical vertebrae. There was no evidence of a cervical rib. Cervical Pantopaque myelography showed a constant defect between the fourth and fifth cervical vertebrae on the right side.

In view of the localized upper limb pain and features suggestive of a scalene syndrome, it was decided to section the anterior scalene muscle and delay surgical attack on the cervical disc until results of scalenotomy had been determined.

The patient was entirely relieved of his complaints by scalenotomy and has been seen in our follow-up clinic at regular intervals over a period of one and three-quarter years since operation.

CASE 4.—J. K., a 40-year-old waxmaker, was admitted to the Graduate Hospital on Nov. 8, 1948. In September, 1947, he had attempted to stop with his left hand a heavy drum of oil rolling down an incline. This abruptly pulled on the left shoulder and caused severe pain in this shoulder and in the left arm. A few weeks after injury he noted numbness of the last three fingers and the ulnar surface of the left forearm.

Prolonged cervical traction gave considerable relief from pain, but with discontinuation of this treatment, the symptoms recurred. On admission he complained of pain in the left shoulder radiating down the lateral aspect of the arm to the elbow and occasionally into the little finger. Sometimes the pain radiated up the left side of the neck and down the left costal cage to the level of the diaphragm.

Examination revealed that there was a fairly full range of neck movements, but all movements except that of the chin to the left were accompanied by the complaint of pain. There was considerable tenderness over the left outer aspect of the neck and over the left shoulder region. Various movements of the left shoulder joint caused pain. There was questionable weakness of the left biceps and triceps and left hand grip. The left triceps reflex was absent.

Except for a total protein content of 75 mg. per cent, the spinal fluid studies were all normal. X-ray examination of the cervical and dorsal spine was essentially negative except for the presence of minimal hypertrophic degenerative changes as indicated by minimal lipping and beak formation about the anterolateral margins of the vertebral bodies. A Pantopaque study of the cervical subarachnoid pathways revealed a constant filling defect on the left, anterior and lateral between the fifth and sixth thoracic vertebrae.

On Nov. 17, 1948, a bilateral cervical laminectomy was performed, removing the laminae of the fourth, fifth, and sixth cervical vertebrae. A definite protrusion of the intervertebral disc between the fifth and sixth cervical vertebrae on the left was identified but found to be too hard to remove. The dentate ligaments were cut and the sixth cervical root thoroughly decompressed, including the opening of the dural root sheath. He was discharged on Nov. 28, 1947, somewhat relieved of his pain.

His pain was less for about six weeks, and radiating pain in the neck and chest never recurred. Following this temporary improvement, he had recurrence of severe pain in the left upper extremity and on Oct. 12, 1949, he was re-explored. The herniated disc between the fifth and sixth cervical vertebrae was again identified. There was no evidence of a cyst or other pathology to suggest an added cause for the return of this man's pain. Following the re-opening of the laminectomy wound the patient continued to complain of constant pain in the left upper extremity.

A left anterior scalenotomy was performed on Dec. 7, 1949. From the first postoperative day until the present, three and one-half months since scalenotomy, the patient has been entirely free of pain.

DISCUSSION

In all four cases reported it is our opinion, although scalenotomy was carried out, that the underlying major etiological factor was a herniation of a cervical intervertebral disc. In three instances there were distinct changes

in the roentgenograms of the cervical spine indicative of traumatic or degenerative change of one or more cervical intervertebral discs, and one of these patients had a confirmatory myelographic study. In the fourth patient, myelographic evidence of a herniated disc was obtained and the lesion confirmed at operation. The relief of pain resulting from scalenotomy, nevertheless, was very impressive in each instance. Spurling and Scoville[1] and Michelsen and Mixter[2] have previously suggested that cervical disc herniations may produce a superimposed scalene syndrome, probably due to spasm of the scalene muscle induced by root irritation. Kristoff and Odom[3] remarked that they have frequently seen evidences of vascular compression, presumably from scalenus spasm secondary to cervical disc lesions. These latter authors, however, specifically stated that the pain in the upper extremity must always be regarded as being of radicular origin and directly due to the disc herniation. None of our patients had vascular symptoms or evidence of vascular compression with any of the standard tests. It can only be concluded that pressure of the scalenus muscle on the brachial plexus was a very important factor in the production of their pain, since section of this muscle gave relief.

Although we were cognizant of the evidences pointing to a cervical disc lesion, complicating factors gave, we felt, sufficient indication for considering scalenotomy. In Case 1 the patient was a known hypertensive with a poor myocardium and at best only a fair risk for cervical laminectomy. Scalenotomy was offered after the failure of cervical traction as a possibly effective treatment less hazardous than cervical laminectomy. In Cases 2 and 3, the patients had persistent, intractable, but not severe pain. The patients refused a major procedure such as laminectomy but were willing to undergo a minor surgical procedure even though its success could in no way be predicted. Scalenotomy was performed in Case 4 only after a laminectomy and a reexploration failed to relieve fully the patient of the upper extremity pain.

From this experience it would seem that there are at least three circumstances in which scalenotomy may be employed in an effort to relieve pain caused by a lateral herniation of a cervical intervertebral disc: (1) in patients who are poor risks for laminectomy; (2) in patients who have residual upper extremity pain following an adequate cervical laminectomy; and (3) as a less formidable procedure than laminectomy in the treatment of minor, but nevertheless intractable, upper extremity pain caused by a cervical disc displacement.

We do not maintain that scalenotomy is effective in every patient, or even the majority of patients. A study of the case records in this series of patients fails to reveal any crucial factors that might be used to determine whether scalenotomy will or will not be successful, except perhaps for tenderness to pressure over the anterior scalene muscle. At this stage of our experience, it can only be stated that in some instances of intractable upper extremity pain, apparently due to herniation of cervical intervertebral discs, scalenotomy may be offered as a therapeutic measure with some hope of suc-

cess when direct attack upon the disc itself is felt to be too formidable a procedure for a particular patient and nonoperative treatment has been ineffective.

SUMMARY

A series of four patients presumably suffering with a lateral herniation of a cervical intervertebral disc and relieved by anterior scalenotomy is reported. It is suggested that the syndrome due to herniation of a cervical intervertebral disc and the scalenus anticus syndrome are not necessarily mutually exclusive. The circumstances in which scalenotomy might be recommended for relief of pain believed due to a herniated cervical disc are discussed.

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VERTEBRAL MANIFESTATIONS OF MALIGNANT LYMPHOMA, MYELOID LEUCEMIA, AND MULTIPLE MYELOMA

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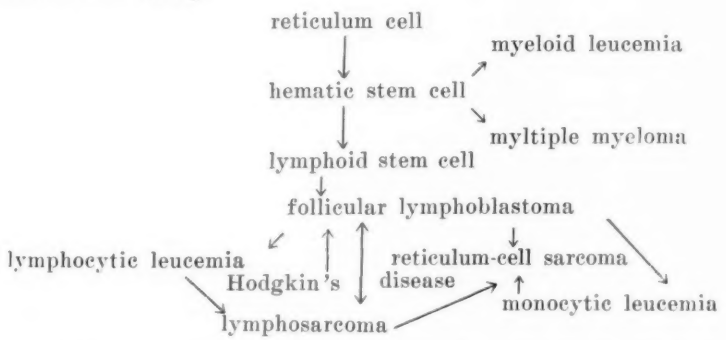
OSSEOUS, particularly vertebral, manifestations of malignant diseases of the reticuloendothelial tissue are more common than is generally supposed. Prior to the advent of roentgenography osseous involvement in these diseases was infrequently recognized. During the past 2 decades skeletal involvement by them has become more generally known, although its frequency and extent are still not fully appreciated. One very compelling reason for this is that the bony involvement is not demonstrated by x-ray films unless there is extensive affection of spongiosa, or destruction of the cortex (Coley,[6] p. 397). The marrow may be almost entirely replaced by tumor tissue without change in the density of the shadow cast in the films. Autopsy studies show that bony involvement invariably is substantially greater than the evidence visible in the films would lead one to believe. Especially in myeloid leucemia, and frequently in multiple myeloma, there is tumor tissue replacement within the bone when no roentgen lesion is demonstrable.

Various classifications of these reticuloendothelial tumors have been advanced by students endeavoring to clarify the existing confusion in nomenclature, origin, and histology of the several entities. Among these are those advanced by Gouski and Holman,[10] Ewing,[8] Pease and McDonald,[15] Haden,[11] and Custer and Bernhard.[7] For the sake of clarity, we have attempted a modification of Custer and Bernhard's diagrammatic classification, and will use that in this study.



Schematic deviation of tumor cells of hematopoietic origin.

The schematic diagram modified from Custer and Bernhard, demonstrates the manner in which the hematic stem cell derives from the reticulum stem cell. From the hematic stem cell both the lymphoid tumors, and the myelogenous tumors develop. Myeloid leucemia and multiple myeloma come directly from this cell, whereas the lymphoma group pass through other transitional stages; namely, the lymphoid stem cell, and the follicular lymphoblastoma.

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The lymphoblastoma can then evolve directly into one of several tumors. These in turn may metamorphose into other types of tumors, as indicated by the arrows on the diagram.

Because a greater amount of investigation has centered about Hodgkin's disease, we have in this study treated Hodgkin's disease separately from the remainder of the malignant lymphoma group. Because multiple myeloma and myeloid leucemia are clinically and histologically separate, and differentiate earlier from the reticulum stem cell, as shown in the diagram, these 2 conditions are also considered separately.

COMMON FACTORS

Before discussing the various clinical entities, a review of the factors common to this entire group of conditions will be made. These diseases, except the acute leucemias of childhood, occur for the most part in the middle decades of life. They are found, roughly, twice as often in males as females. They are systemic diseases with variable symptom complexes and clinical pictures. The symptoms, signs, and laboratory findings are in a large measure dependent upon the organs involved. Bone involvement is an early, rather than a late, manifestation of these conditions. There are no pathognomonic roentgen changes. The films more often show osteolytic, but may disclose osteoblastic or periosteal, reactive bony lesions. They may suggest fibrous dysplasia, bone cysts, osteomyelitis, osteogenic sarcoma, Ewing's tumor, or metastatic carcinoma. Pathologic fracture may occur. The prognosis for all is poor, but is a little better in older patients. In general, the treatment is palliative and symptomatic for visceral lesions. X-ray therapy is helpful. Recent reports on the use of nitrogen mustard and radioactive phosphorus are encouraging, except in Hodgkin's disease and multiple myeloma (Low-Beer and associates,[14] and Hempelmann and colleagues[12]). Where a solitary tumor exists in an accessible spot surgery is indicated.

Table I is a recapitulation of our study based on 62 autopsies, divided as follows: Hodgkin's disease 28, lymphosarcoma 8, reticulum-cell sarcoma 6, lymphatic leucemia 5, multiple myeloma 8, and myeloid leucemia 7.

X-ray studies disclosed vertebral lesions in 1 case of Hodgkin's disease, 1 case of reticulum-cell sarcoma, 3 cases of multiple myeloma, and 1 case of myeloid leucemia.

X-ray involvement was noted in bones other than vertebral column as follows: in the Hodgkin's group, 4; in the multiple myeloma group, 4; and in the myeloid leucemia group, 1. X-ray lesions were disclosed in 14.5 per cent.

At autopsy it will be noted that the number of cases showing bony involvement is substantially higher. In the Hodgkin's disease group 20 showed involvement of vertebrae, and 12 involvement of other bones. Actually, we believe that 21 showed vertebral involvement, but unfortunately one autopsy protocol did not specify in which bone the lesion was found. It has been our experience that when any other bone is involved the vertebral column invariably shows changes. This table shows that the same condition obtains in regard to

the other diseases studied. The percentage of vertebral involvement in these various groups is shown in the next to last column on the right, 71.4, 75, 66.6, 80, 87.5, and 100. If we accept the figure of 20 in the Hodgkin's disease group, the percentage showing vertebral involvement for the entire series is 77.4. However, if one concedes that in 21 of these patients the vertebrae were involved, the percentage for the group is 79.

TABLE I.

NUMBER OF CASES AUTOPSIED	X-RAY LESIONS		AUTOPSY LESIONS		PERCENTAGE VERTEBRAL LESIONS	ASSOCIATED LESIONS
	VERTEBRAE	OTHER BONES	VERTEBRAE	OTHER BONES		
			Hodgkin's disease			
28	1	4	20 (21?)	12	71.4 (75?)	1 pathologic fractured rib 1 case with rib lesion, vertebrae not mentioned
			Lymphosarcoma			
8	0	0	6	2	75	
			Reticulum-cell sarcoma			
6	1	0	4	0	66.6	1 Mycosis fungoides (? Hodgkin's)
			Lymphatic leucemia			
5	0	0	4	0	80	1 patient died of carcinoma of prostate with metastases 1 patient died of miliary tuberculosis
			Multiple myeloma			
8	3	4	7	6	87.5	3 compression fractures of spine 1 pathologic fracture of femur
			Myeloid leucemia			
7	1 with sclerosis of vertebrae and pelvis		7	3	100	
Totals						
62	6	9	48	23	77.4 (Corrected 79)	

It is observed that one of the Hodgkin's patients sustained a pathologic rib fracture. In the lymphatic leucemias concomitant diseases are often encountered, and 2 of our patients actually died from other causes. In the multiple myeloma group, 3 of the 7 cases which demonstrated vertebral involvement had compression fractures of the spine. One of these patients also had a pathologic fracture of the femur.

Table II depicts further details of this study.

Only those cases which exhibited bony lesions at autopsy are included here. Of these 49, only 6 showed definite vertebral lesions by x-ray while living, and 9 lesions of other bones. It will be observed, however, that these figures may be in error, since in 6 instances no vertebral films and in 11 instances no films of other bones were made.

Similarly, it is probable that bones other than the vertebrae were involved in more than the 23 cases shown here, as in an equal number no other bones were

mentioned in the autopsy protocols. Steiner[16] has shown quite conclusively that the greater the number of bones examined, the larger the percentage of cases in which bony lesions can be demonstrated.

In the last column the ages are tabulated. The first figures are the ages of the youngest patient in each group, the second, the ages of the oldest. The figures in parentheses are the average ages in each group.

TABLE II.

NUMBER OF CASES	VERTEBRAL LESIONS		OTHER BONES		AUTOPSY LESIONS		OTHERS NOT MENTIONED	
	X-RAY	NO FILM	X-RAY	NO FILM	VERTEBRAE	OTHER BONES		
				Hodgkin's disease				Ages 26-77 (40)
21	1	3	4	6	20 (21?)	12	9	Biopsies 15
								Vertebral biopsy 1
				Lymphosarcoma				Ages 21-58 (47)
6	0	0	0	1	6	2	3	Biopsies 3
								Sternal aspiration 1
				Reticulum-cell sarcoma				Ages 22-55 (37)
4	1	1	0	1	4	0	4	
				Lymphatic leucemia				Ages 25-83 (60)
4	0	1	0	2	4	0	2	
				Multiple myeloma				Ages 44-70 (57)
7	3	0	4	0	7	6	1	Bence Jones protein +2 −4
	1 marked osteoporosis of vertebrae and pelvis							A/G ratio reversed 3
								Hypercalcemia 2
								Vertebral biopsy 1
								Sternal aspiration 5
				Myeloid leucemia				Ages 24-58 (48)
7	0	1	1	1	7	3	4	Sternal aspiration 3
49	6	6	9	11	48	23	23	Average age 45.6

The number of biopsies (18) and sternal aspirations (9) done as diagnostic procedures are here shown. In 2 instances, 1 Hodgkin's disease, and 1 multiple myeloma, the diagnosis was established by vertebral biopsy. Of the 7 cases of multiple myeloma, the Bence-Jones protein was positive in 2 cases, negative in 4 cases, and not recorded in 1. There was reversal of the A/G ratio in 3 instances, and hypercalcemia in 2.

We cannot prove, but we believe, from a study of the literature and these cases, that bone involvement in this group of diseases is frequently an early rather than a late occurrence.

A detailed discussion of each of the conditions previously mentioned is not possible in this short presentation. Several illustrations pertaining to Hodgkin's disease are presented, as well as 3 illustrating some typical changes in the multiple myeloma group.

HODGKIN'S DISEASE

Hodgkin's disease, as previously stated, is a disease affecting all the lymphatic tissues of the body. Numerous authors have noted that those

organs which retain much reticuloendothelial tissue, namely, lymph nodes, liver, spleen, and bone marrow, are commonly involved in this condition. Those organs containing relatively little reticuloendothelial tissue, namely, tonsils, thymus, and lymph tissue of the gut, are involved with less frequency.

Collected Data.—Table III re-illustrates that bone lesions in Hodgkin's disease are demonstrated much more frequently at autopsy than clinically or by x-ray. Steiner[16] collected from the literature and from his own material 2,006 cases of Hodgkin's disease, of which clinically 8.3 per cent evinced bone lesions. Of these 547 came to autopsy. The pathologists discovered that of this number 28.3 per cent exhibited osseous lesions. Falconer and Leonard[9] compared the frequency with which bone lesions could be demonstrated by x-ray and by post-mortem examination. Of 750 cases studied roentgenographically, they discovered that 14.9 per cent showed definite bone involvement. At autopsy 53.3 per cent of 213 patients gave evidence of disease of the bones. In our much smaller series the percentage in which films show bony lesions is not dissimilar from those of previous series, namely, 17.8. At autopsy, however, 71.4 per cent of the cases showed osseous involvement. This higher percentage than is commonly reported is undoubtedly due to the fact that our pathologists were seeking bony lesions.

TABLE III. COLLECTED DATA ON FREQUENCY OF BONE LESIONS IN HODGKIN'S DISEASE

NUMBER OF CASES	PER CENT WITH BONE LESIONS	NUMBER OF CASES AT AUTOPSY	PER CENT WITH BONE LESIONS (AUTOPSY)
Steiner (Studied clinically)			
2,006	8.3	547	28.3
Falconer and Leonard (Studied by x-ray)			
750	14.9	213	53.3
Present series (Studied by x-ray)			
28	17.8	28	71.4

Fig. 1 is a roentgenogram of several vertebrae, a sternum, and 4 ribs, which were removed from a patient with Hodgkin's disease. It will be seen that there is no gross evidence of pathology (the irregularities present here were made by the autopsy saw).

Fig. 2 shows a photograph of these same bones. It is seen that the medulla of all bones is practically entirely replaced by tumor tissue. This illustrates the statement so often made that there may be extensive skeletal involvement, but unless the cortex is destroyed, or collapse occurs, roentgen evidence of this involvement is not present.

Fig. 3 is a microscopic section of one of these vertebrae in which the characteristic Reed-Sternberg cells and eosinophils are found.

The following slides are from a rather advanced case of multiple myeloma. Fig. 4 shows the lytic lesions with thinning of the cortices in the femur and humerus. Fig. 5 shows the marked osteoporosis of the vertebrae, pelvis, and

upper femora, characteristic of the later stages of multiple myeloma. Pathologic fracture of the femoral neck is also seen. Fig. 6 is a slide of this case showing the typical closely packed myeloma cells of uniform size.

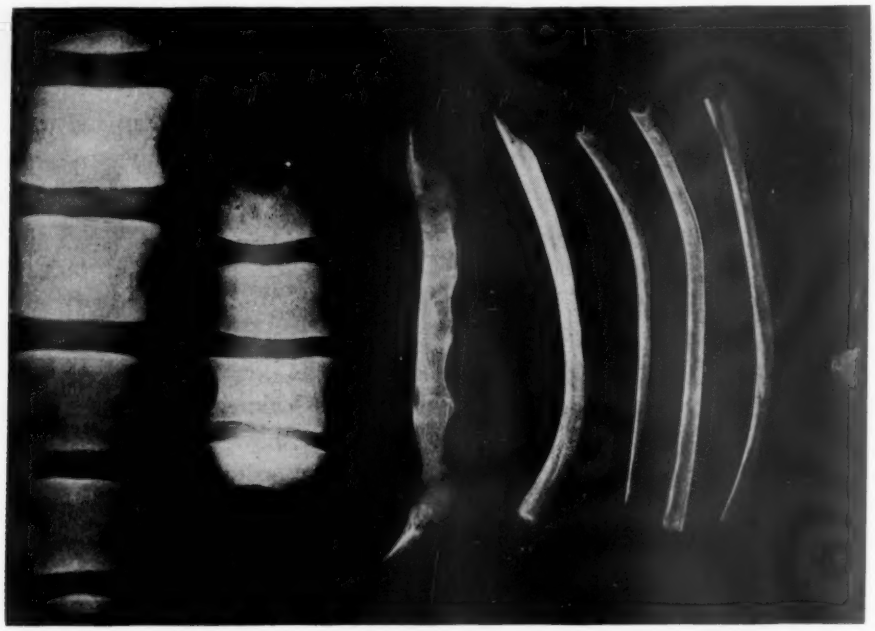

Fig. 1.

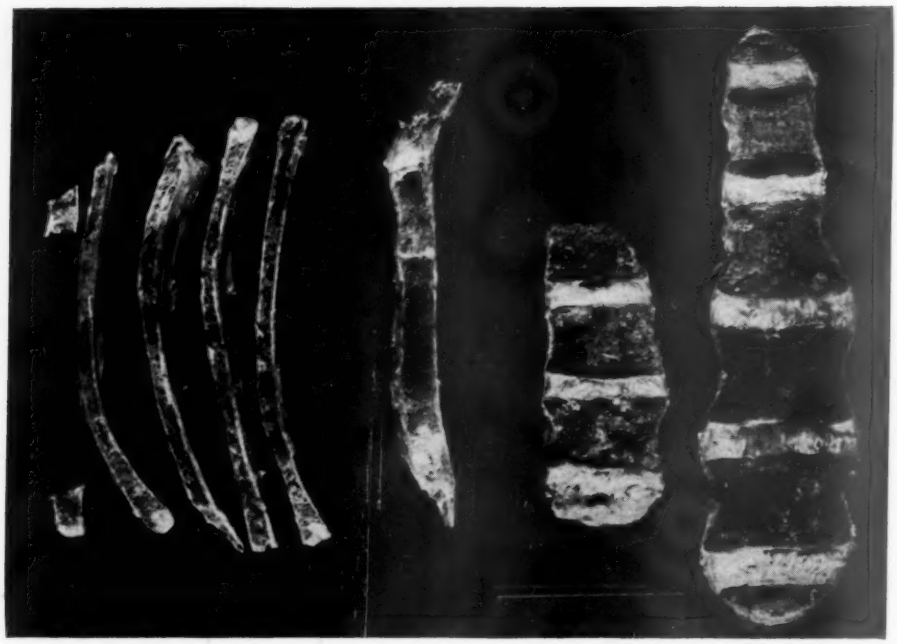

Fig. 2.

Before concluding I should like to mention a little used means of establishing a diagnosis which has been helpful to us in several instances and conditions. In 18 of our cases the diagnosis was made by sternal aspiration, in 9

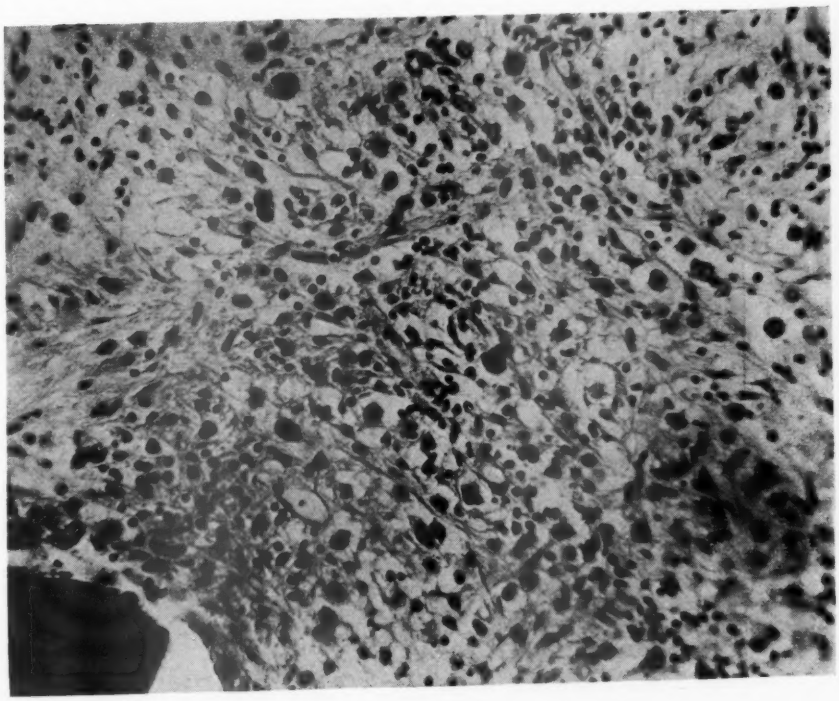

Fig. 3.

A. *B.*

Fig. 4.

by lymph node biopsy. The value of these procedures cannot be overemphasized (Aegerter and Robbins,[1] Coley,[6] Bayrd,[2] Beizer and associates,[3] Bethell,[4] Lichtenstein and Jaffe,[13] and Bichel and Kirketerp[5]).

Fig. 5.

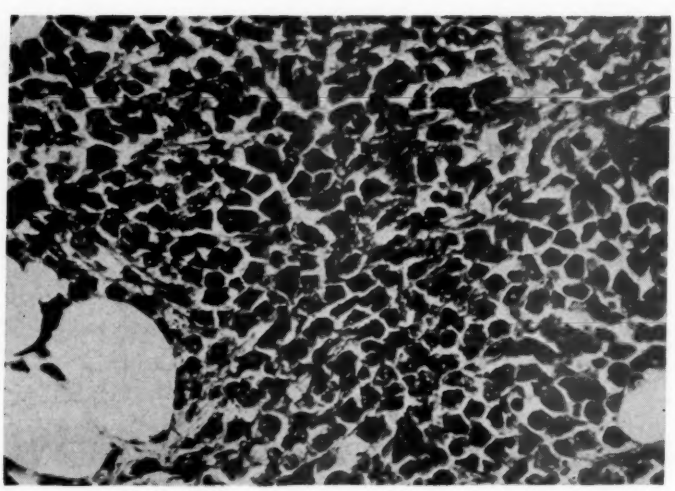

Fig. 6.

We have recently been performing vertebral biopsies in vertebral lesions of questionable nature. The diagnosis in 2 cases in this series was made by this procedure. It has proved extremely valuable in a number of conditions. Our technique is essentially the same as that of Jose Valls and associates.[17]

Fig. 7.

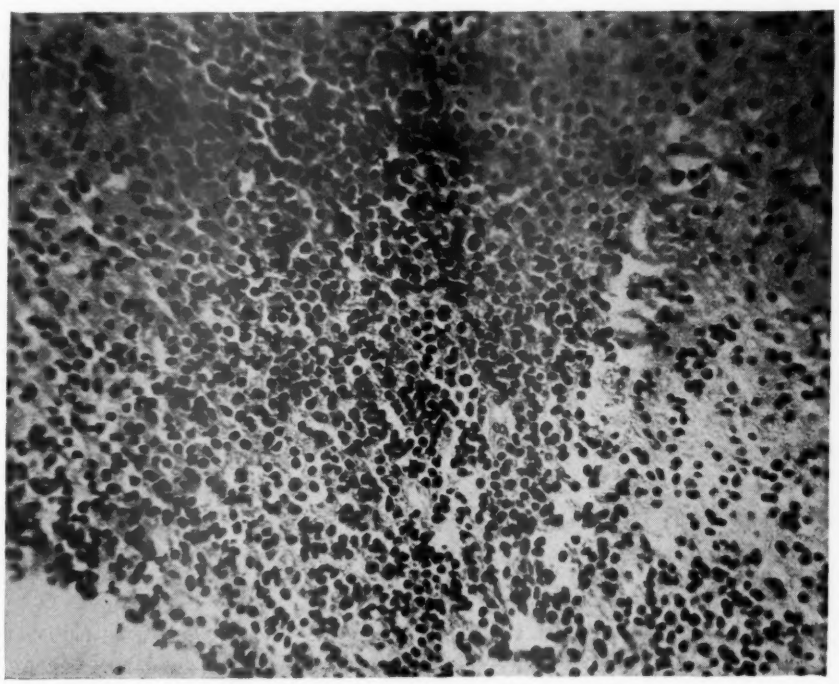

Fig. 8.

Fig. 7 shows the destructive lesion of the body of the third lumbar vertebra. The biopsy needle is observed entering the lesion.

Fig. 8 shows a photomicrograph of the biopsy specimen disclosing a malignant lymphoma. The precise type cannot be surely determined from this slide. It proved to be Hodgkin's disease.

CONCLUSIONS

1. This study has demonstrated that osseous lesions, particularly vertebral lesions, are not uncommon in the diseases of the malignant lymphoma group. They are considerably more frequent and more extensive and we believe they occur earlier in the course of these diseases, than is generally realized.

2. X-ray studies reflect this involvement only if there has been destruction of the cortex or collapse.

3. Biopsy of an involved vertebral body is sometimes helpful in arriving at a diagnosis.

Acknowledgment and thanks to the Department of Pathology, and in particular Dr. Louis Lichtenstein, for aid in the preparation of this paper are hereby tendered.

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TENODESIS OF BICEPS TENDONS

A Preliminary Report

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THE purpose of this paper is to present a method of tenodesis of ruptured biceps tendons which permits better control of the tension of the resutured muscle, expedites operative and convalescent time, and also gives excellent security of the tendons.

Rupture of the biceps tendon is a comparatively frequent lesion encountered at the United States Marine Hospital, Staten Island, N. Y., due to common occurrences among mail handlers in the large post offices in New York City. The history of the injured patient usually states that catching heavy mailbags coming down chutes precipitated the lesion. However, it is not our purpose to discuss the pathogenesis, pathology, or diagnosis of this lesion. Steindler has given an excellent discussion as to these, and we have only one statement to add: our patients have usually been active, healthy men 35 to 50 years of age who have not shown gross degenerative changes in the tendons or previously suffered from abnormal conditions in the shoulder or elbow.

It has been the policy of our orthopedic department to explore all ruptures or tears, whether complete or incomplete, as past experience has taught us that conservative treatment of partial tears is too uncertain and, usually, the final disability is such that operative intervention is indicated.

There are several methods of surgical repair described in texts. Whenever the long head is ruptured near the glenoid lip, leaving a long tendon to be sutured somewhere, it has been recommended that the tendon be attached to the coracoid process or sutured to the short head of the biceps. It has also been recommended that two drill holes be placed through the humerus in a coronal plane above the deltoid insertion and that the long head tendon be threaded through this hole and sutured back on itself. This procedure requires wide exposure to permit drilling and further procedures which endanger the shaft strength of the humerus. End-to-end suture has been found satisfactory in some cases of long head ruptures when they occur in the distal half of the tendon. Sutures to the pectoralis major muscle or to the intertubercular sulcus, as recommended by some authors, have not been used in our clinic.

As to ruptures of the radial insertion of the biceps, Bunnell has devised a procedure for suturing the ruptured distal tendon with stainless steel wire of the pull-out type back to the biceps tuberosity after chipping up its surface.

Presented as part of an exhibit at the meeting of the American Academy of Orthopedic Surgeons, Waldorf-Astoria Hotel, New York, N. Y., February, 1950.

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The insecurity of coaptation of a broad base tendon with a limited chipped area leaves much to be desired by use of the pull-out wire. Ruptures of the distal tendon are considerably less frequent than the long head ruptures.

The two situations which have stimulated us to devise the "key slot" tenodesis are, first, those cases which have tears proximal to or within the bicipital groove and, second, those old cases with musculotendinous gap and wide scarring where grafting was necessary, using the biceps tendon itelf as a graft if necessary. The method has also been of great value in reviewing the painful snapping biceps syndrome.

METHODS

Two locking procedures are described in tenodesing proximal and distal tendons of the biceps. The first will be referred to as the "key slot" method, the latter, the "trap-door" slot method (Fig. 1).

In our experience, the key slot method has been the most successful for the long head tendon repairs, whereas the trap-door slot method, using a small oscillating blade to prepare the window, is quite advantageous in the repair of frayed biceps disinsertions.

In either case the tendon is first exposed through an anterolateral incision and prepared for tenodesis. In the case of tears of the long head of the biceps, this preparation is accomplished by either sewing the end back on itself or tying the end of the tendon in a knot and suturing or constructing the end of the tendon into a ball by multiple cotton sutures as illustrated. The adherent or retracted muscle belly is then freed and stretched to what is judged to be its normal length and the site where the key slot is to be made is marked on the anterior surface of the humerus. First, a three-sixteenth inch drill is used to make a hole proximal to the mark; then a second, one-eighth inch, drill hole is made distally, at a distance of one-eighth inch from the first hole. Next a dental chisel or small osteotome is used to open a narrow slot between the two holes, making a single hole shaped similar to a keyhole slot. Placing the bulbous portion of the previously prepared tendon into a large hole and drawing it down into the slot locks it firmly into the groove and small hole and holds the muscle at the tension previously calculated as correct.

In the trap-door slot method, which is preferred in repair of the frayed disinsertions of the biceps, the biceps tuberosity of the radius is exposed by the usual method. Then, a Luck saw or similar oscillating small blade is used to cut a rectangular block around the tuberosity, leaving the proximal one-third of the tuberosity intact and going about one-eighth inch distally on the shaft. The block is removed by prying with an osteotome; then a notch is rongeured out in semicircular fashion. Also, a semicircle on the remaining tuberosity is rongeured out, completing the circle for the reinsertion of the tendon as illustrated. This hole is smaller than the diameter of the tendon in order that the tendon may be solidly fixed by the block, as a keystone wedge, holding the tendon firmly. It is not necessary to suture the tendon down or use pull-out wires. Early motion is instituted.

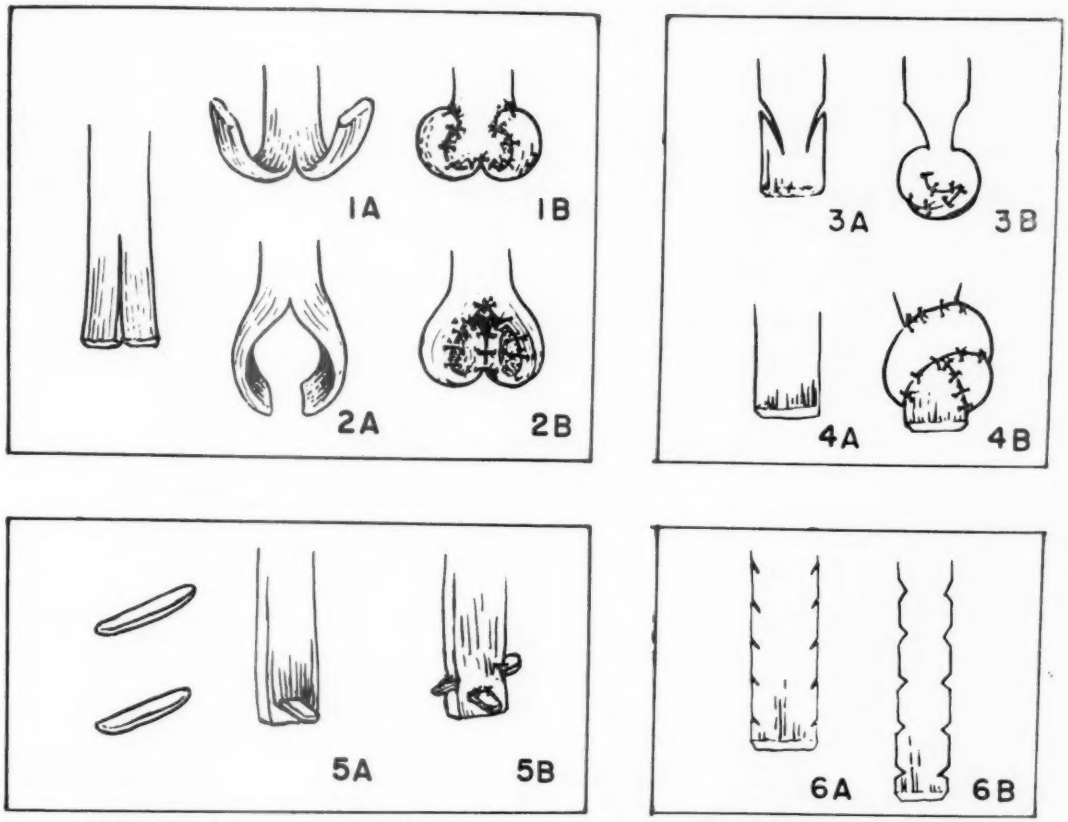

Fig. 1.—The preparation of the avulsed tendon is illustrated. The method of locking the tendon in the key slot on the disinserted biceps tendon distally is shown.

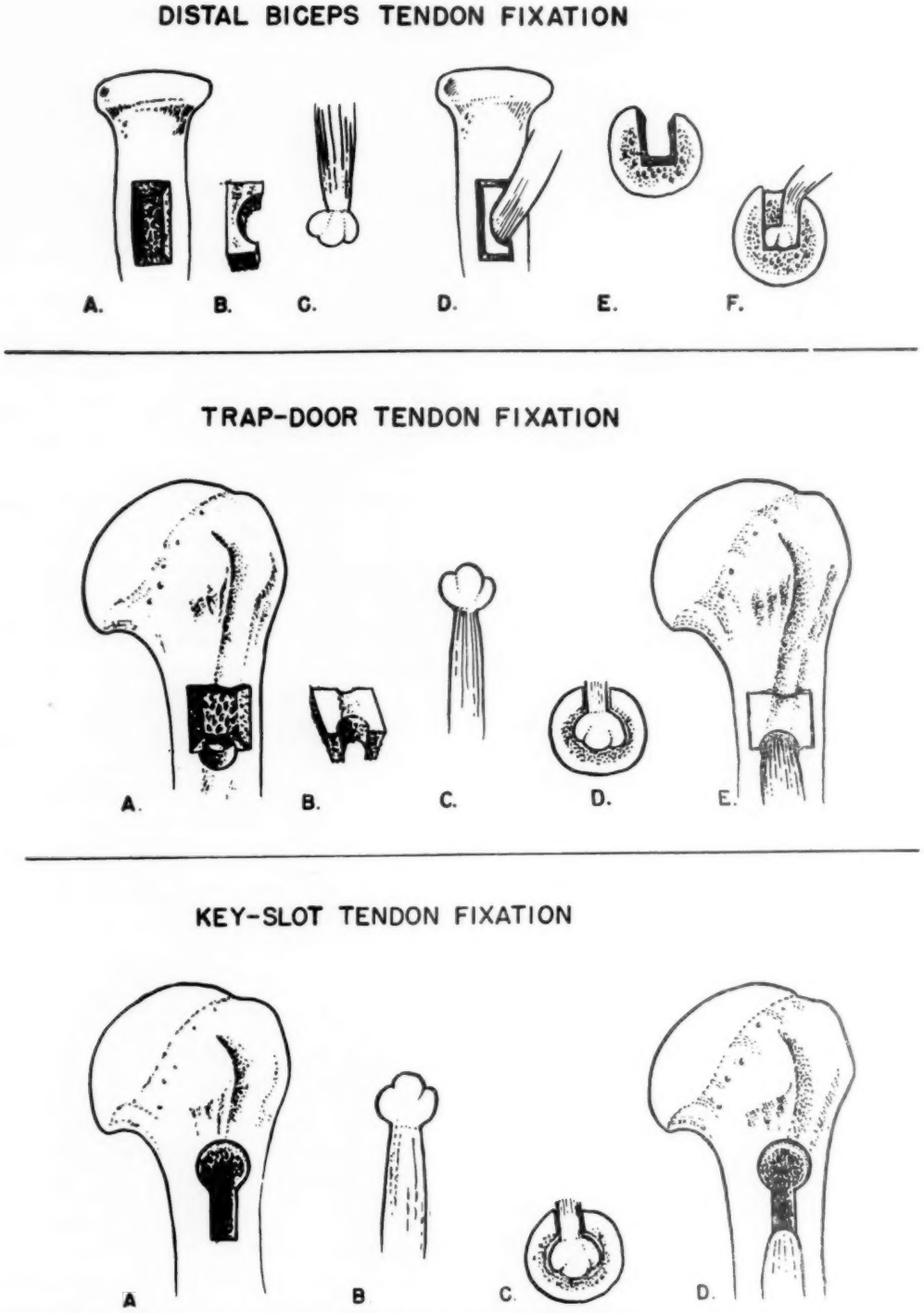

Fig. 2.—The method of locking the biceps tendon by the trap door is shown. Also the key slot fixation is illustrated. The key slot is the most rapid procedure and probably provides more solid fixation.

COMMENT

It has been particularly gratifying to have patients begin active motion using the repaired muscle within a day or two after surgery. Very little, if any, pain has followed surgery in our cases, so that physical medicine could usually be instituted the first postoperative day. There has been minimal edema postoperatively in comparison to some of the older methods and the cephalic vein can be more readily spared in long head tendon repairs.

SUMMARY

Methods of tenodesis of biceps tendons have been presented. These have been described as "key slot" and "trap-door slot" methods. The procedures and illustrations of the methods are presented.

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EXPERIMENTAL ARTERIOVENOUS FISTULA: HISTOLOGIC CHANGES IN THE SMALL COLLATERAL ARTERIES

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CLINICAL experience with chronic vascular insufficiency in lower extremities from which arteriovenous fistulas had been excised prompted investigation of that problem in the experimental animal.[1] In a previous publication[2] roentgenographic demonstration of regression of the collateral arterial bed after fistulectomy was presented, and certain pathologic changes in the smaller collateral arteries were briefly mentioned. These lesions supplied evidence of an organic change in the collateral vessels which seemed to be important in understanding the status of the vascular supply in an extremity after fistulectomy. Further investigation now permits us to describe these changes in more detail. No report of similar observations on such vessels has been found in the literature.

The material utilized in this investigation was obtained from 11 dogs with femoral arteriovenous fistulas. In 9 dogs the fistulas were excised after 11 to 39 weeks and the animals were maintained for intervals of 2½ to 18 weeks thereafter. Two other dogs without fistulectomy were sacrificed 14 and 36 weeks, respectively, after construction of the fistula.

EXPERIMENTAL RESULTS

Pathologic changes were observed in arteries of the collateral bed that ranged from 75 to 1,200 microns in outside diameter. The most severely affected vessels were those between 200 and 500 microns. Except in the immediate region of the fistula the larger arteries such as the iliac and femoral and their larger branches failed to show significant changes. The veins of the collateral bed exhibited no changes similar to those in the small arteries and except for slight thickening of their walls did not appear significantly altered. In many sections a slight to moderate degree of arteriolar hypertrophy was noted, but the experiments as designed did not permit complete evaluation of these changes. Arteriolar hypertrophy might indicate a sustained arteriolar contraction as an attempt to restore the peripheral resistance in the region of the arteriovenous shunt. The changes in the arterioles, however, were distinctly different in type and extent from the lesions in the arteries.

In the affected small arteries the pathologic process involved principally the internal elastic membrane and the subendothelial tissue of the intima. The internal elastic membrane was fragmented, laminated, frayed, and sometimes partially or even totally destroyed. The intima contained increased amounts of subendothelial tissue which varied from a few cells in thickness to a wide zone

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which almost obliterated the lumen. Although this zone varied in depth around the internal circumference of the artery, it was never focal or plaquelike. The cells of this tissue had a fusiform shape and contained small ovoid nuclei. Their identity as fibroblasts in the more advanced lesions was established by staining collagenous fibers between the cells with the Van Gieson and Masson trichrome stains. In arteries with an advanced proliferation of subendothelial fibrous tissue the internal elastic membrane was usually reduced to a few short fragments or totally destroyed. Less advanced stages of the 2 lesions could not be consistently correlated. In arteries with minimal to moderate fibrous lesions of the intima the Verhoeff elastic tissue stain sometimes revealed fine elastic fibrils and a black smudgy appearance in the inner portion of the subendothelial zone. In the most advanced fibrous lesions no traces of elastic fibrils were present except for occasional fragments of the original internal elastic membrane. The media and adventitia were only rarely affected, but occasionally new fibrous tissue extended from the lesion in the intima into the inner layers of the media. A thickening of the adventitia was present in a few specimens.

The distribution of affected arteries was inconstant even in the same block of tissue. In many sections uninvolved vessels lay in close proximity to involved vessels of the same or of different sizes. However, in any section where advanced changes were present in some arteries most of the remaining arteries showed moderate changes. No recent or organized thrombi were encountered. The lumen was sometimes reduced to a small fraction of the total diameter of the vessel by proliferation of subendothelial tissue. In most instances an accurate appraisal of the reduction in the lumen was impossible, since the degree of dilatation and contraction of the vessel in the pre- and postfistulectomy periods could not be determined. In no instance was complete obliteration of the lumen observed.

Wide variation in the development of the collateral circulation was encountered despite efforts to create a standard experimental preparation. Factors causing this variation have been discussed previously.[2] In one-half of the animals, roentgenographic studies made immediately after fistulectomy revealed the extent of the arterial collaterals.* In these and in the remaining animals an estimate was also made at the time of fistulectomy. The persistence of a strong thrill and marked distal pulsation after proximal occlusion of the artery correlated well with an extensive collateral bed. The degree of dilatation of the distal artery roughly indicated the volume of retrograde blood flow into the fistula. The importance of the distal artery in determining the development of the collateral circulation has recently been pointed out by Holman[3] and has also been noted in our investigations.[2]

By utilizing such observations, an attempt was made to correlate the degree of the pathologic lesion with the development of the collateral bed. The incidence and magnitude of the arterial lesions were arbitrarily graded and the results compared with the estimate previously made of the collateral circulation. In

*For these studies 75 per cent Neo-iopax was supplied by the Schering Corporation, Bloomfield, N. J., and 70 per cent Diodrast by the Winthrop Chemical Company, Inc., New York, N. Y.

Fig. 1. Fig. 2.

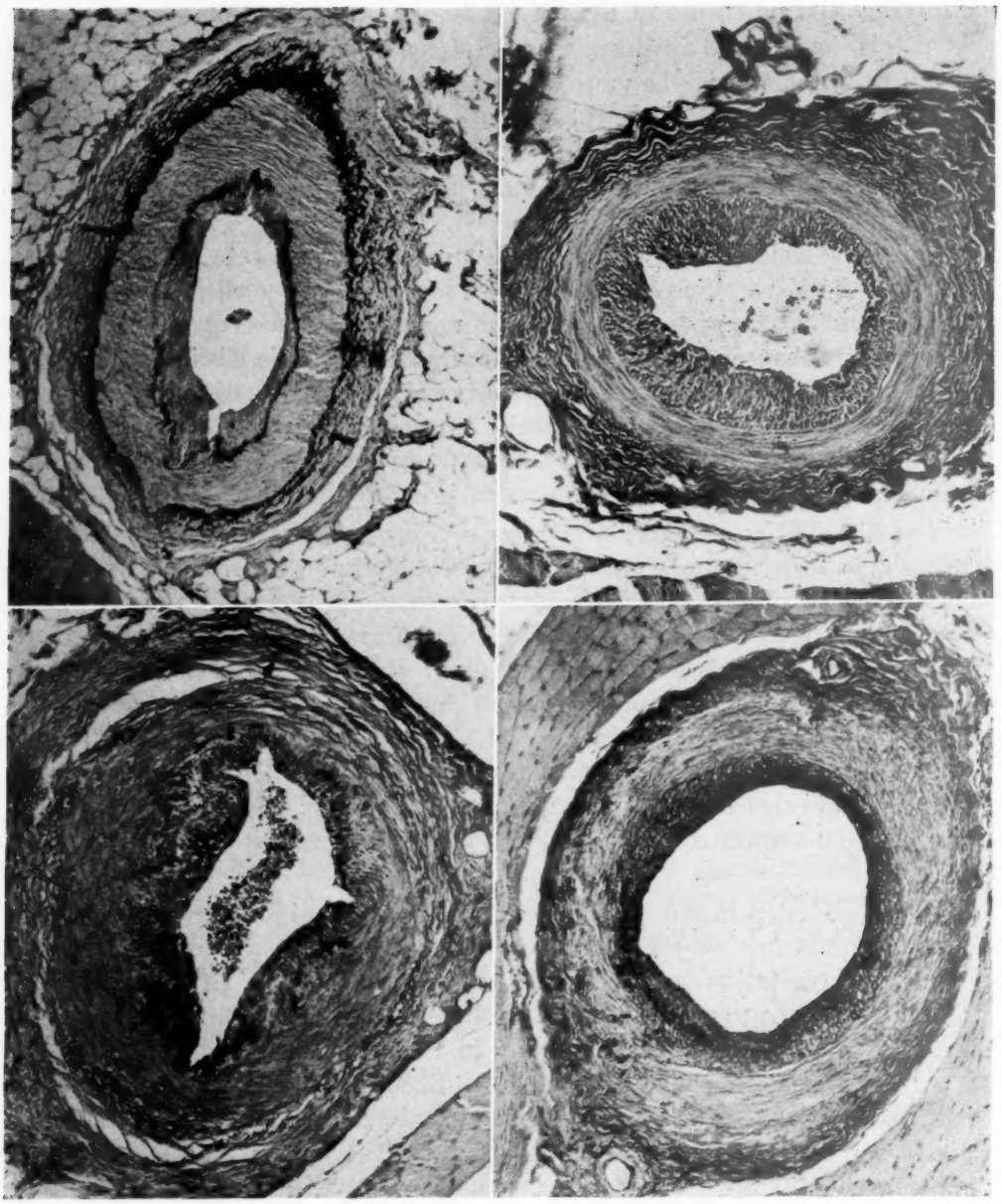

Fig. 3. Fig. 4.

Fig. 1.—Moderate intimal hyperplasia and fragmentation of internal elastic membrane. Outside diameter of artery; 630 microns. Duration of fistula, 15 weeks. Sacrificed 7 weeks postfistulectomy (Verhoeff-Van Gieson stain, ×70).

Fig. 2.—Moderate intimal hyperplasia. Outside diameter of artery, 330 microns. Duration of fistula, 14 weeks. Sacrificed 7 weeks postfistulectomy (hematoxylin and eosin stain, ×150).

Fig. 3.—Moderate intimal hyperplasia and marked fragmentation of internal elastic membrane. Outside diameter of artery, 600 microns. Duration of fistula, 18 weeks. Sacrificed 5 weeks postfistulectomy (Verhoeff-Van Gieson stain, ×105).

Fig. 4.—Lamination of internal elastic membrane and fine elastic fibrils in intima. Outside diameter of artery, 480 microns. Duration of fistula, 19 weeks. Sacrificed 5 weeks postfistulectomy. Relatively large lumina of vessels persisting in this animal were possibly related to thrombosis of proximal artery found at fistulectomy (Verhoeff-Van Gieson stain, ×115).

Fig. 5. Fig. 6.

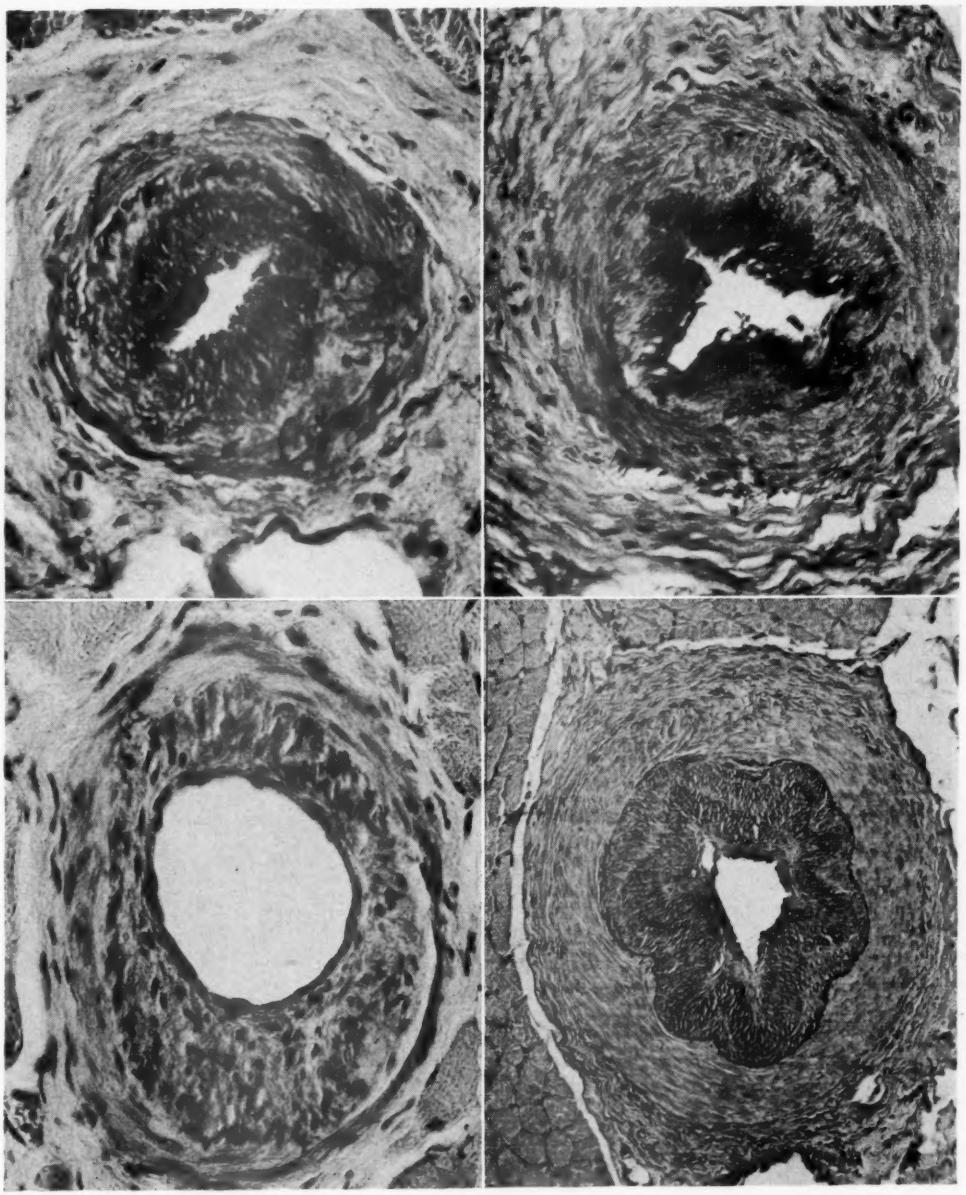

Fig. 7. Fig. 8.

Fig. 5.—Marked intimal hyperplasia. Fragmentation and fraying of internal elastic membrane. Outside diameter of artery, 110 microns. Duration of fistula, 16 weeks. Sacrificed 2½ weeks postfistulectomy (Verhoeff-Van Gieson stain, ×400).

Fig. 6.—Marked intimal hyperplasia. Specifically stained fine fibrillary and amorphous material in inner zone of intima. Outside diameter of artery, 250 microns. From same dog as in Fig. 2 (Verhoeff-Van Gieson stain, ×240).

Fig. 7.—Moderate intimal hyperplasia. Almost complete destruction of internal elastic membrane. No proliferation of elastic tissue. Outside diameter of artery, 120 microns. Duration of fistula, 11 weeks. Sacrificed 10 weeks postfistulectomy (Verhoeff-Van Gieson stain, ×380).

Fig. 8.—Advanced intimal fibrous proliferation with slight destruction of internal elastic membrane. Outside diameter of artery, 510 microns. Duration of fistula, 17 weeks. Sacrificed 7 weeks postfistulectomy (Verhoeff-Van Gieson stain, ×105).

Fig. 9. Fig. 10.

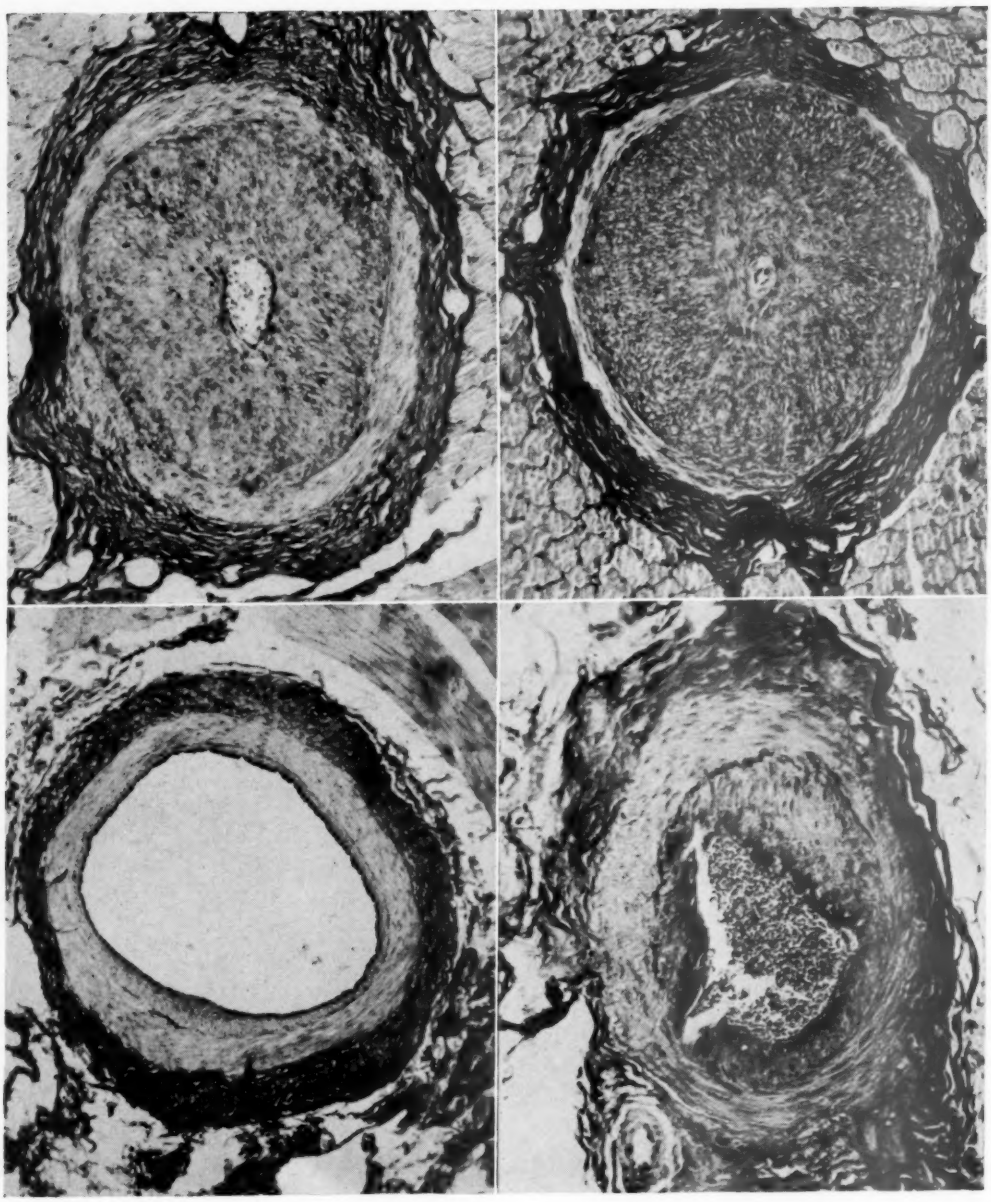

Fig. 11. Fig. 12.

Fig. 9.—Advanced intimal fibrous proliferation with complete destruction of internal elastic membrane. Outside diameter of artery, 420 microns. From same dog as in Fig. 1 (Verhoeff with phloxine stain, ×130).

Fig. 10.—Extreme intimal fibrous proliferation. This artery was most nearly obliterated of any observed. Outside diameter of artery, 530 microns. From same dog as in Figs. 1 and 9 (Verhoeff with phloxine stain, ×112).

Fig. 11.—Moderate intimal hyperplasia and fragmentation of internal elastic membrane. Note dilatation of lumen. Dog sacrificed without excision of fistula. Outside diameter of artery, 1,100 microns. Duration of fistula, 15 weeks (Verhoeff-Van Gieson stain, ×50).

Fig. 12.—Moderately advanced lesion in same dog as in Fig. 11. Outside diameter of artery, 300 microns (Verhoeff-Van Gieson stain, ×200).

general, those dogs with the most extensive collateral circulation at the time of fistulectomy showed the most advanced arterial lesions on subsequent examination. Furthermore, the more extensive lesions were found in the midthigh in the medial and posterior muscle groups where the maximal vascular development was demonstrated roentgenographically. Minimal changes only were found in animals with poorly developed collateral circulation. No significant lesions were observed in specimens taken from the muscles of the calf, where roentgenographic studies failed to reveal collateral vessels.

In the group of dogs sacrificed at intervals after fistulectomy the period of time between fistulectomy and sacrifice could not be correlated with either the number or intensity of the lesions. One dog which had excellent collateral circulation and was sacrificed 2½ weeks after fistulectomy had as many and as extensive lesions as any other animal with the exception of 2 dogs which were sacrificed 6 weeks after fistulectomy. On the other hand, a dog that was not sacrificed until 18 weeks after fistulectomy had a poorly developed collateral bed at fistulectomy and very few well-developed lesions in the arteries. The histologic studies indicated that the arterial lesions were well developed as early as 2½ weeks after fistulectomy and that no notable progression occurred beyond 6 weeks postfistulectomy. Studies by serial biopsies in the same animal were not attempted because it was anticipated that there would be difficulty in obtaining comparable specimens and that the biopsy itself would influence the collateral bed and perhaps induce pathologic changes.

The influence of the fistulectomy and regression of the collateral bed on the morphologic lesions was studied by comparison with the material from the 2 dogs that were sacrificed without excision of their fistulas. These animals, with arteriovenous fistulas of 14 and 36 weeks duration, respectively, had developed excellent collateral circulation. Each had been prepared with bilateral lumbar sympathectomy, but there was no evidence to suggest that the sympathectomy had altered the pathologic picture in the smaller arteries other than by increasing the degree of dilatation of the vessels. Definite arterial lesions similar to those already described were present in each animal on the side of the fistula. The degree of fibrous proliferation was difficult to estimate because of the extreme dilatation of the vessels, but the lesions were less advanced than those from dogs with a comparable collateral circulation sacrificed during the postfistulectomy period. In addition, well-developed lesions have been observed in another dog without fistulectomy and without sympathectomy. In this animal, from another experimental group, an iliac arteriovenous fistula had been present for 1 year. Evidence of the development of the morphologic changes in the walls of the arteries prior to excision of the fistula also was supplied by histologic study of the specimens removed from 2 dogs at fistulectomy. In each there were definite examples of the lesion in small arteries in the tissue about the excised fistula.

In many of the animals ligation of the femoral artery and vein in the contralateral leg had been performed at the time of constructing the fistula. In some instances minimal arterial lesions were observed in the collateral vessels on

the contralateral ligated side. A moderate dilatation of these collateral vessels could be demonstrated roentgenographically, and the morphologic changes were interpreted as being related to the increased blood flow around the obstructed main artery.

The radiopaque media used in some of these experiments to demonstrate the extent of the collateral bed had no effect on the development of the lesions in the arteries. In one-half of the animals these materials were not used, and in control animals with intact circulation no arterial changes developed after arteriography.

In almost all histologic sections examined the arteries appeared more numerous on the side of the fistulectomy as compared to the contralateral intact or ligated side. Since arteriography indicated a nearly complete regression of collateral vessels after fistulectomy in most animals, it is apparent that roentgenographic visualization of the smallest collateral vessels was not obtained.

DISCUSSION

The observations in these experimental animals clearly establish that a degenerative lesion develops in the small arteries of the collateral bed around an arteriovenous fistula in dogs. Arterial lesions of the type described are not naturally occurring phenomena in normal or aged dogs, as they have not been found in control studies of animals with intact circulation to the hind limbs. Several authors[4, 5] have described changes occurring in the immediate vicinity of the fistula, namely, the "venafication" of the proximal artery and the "arterialization" of the associated vein. In studying experimental femoral fistulas, Fick,[6] however, noted the absence of histologic changes in the iliac artery proximal to the fistula. None of these authors mentioned the lesions in the collateral arteries described in this report. Liebow and associates[7] have recently described subendothelial fibrous proliferation and fragmentation of the internal elastic membrane in collateral bronchial arteries after ligation of the pulmonary artery.

Two dogs with excellent collateral circulation showed only moderately advanced lesions when sacrificed without excision of the fistulas. Other animals, even some with less well-developed collateral circulation, showed more advanced lesions when sacrificed during the postfistulectomy period. Advanced lesions were observed in 1 animal 2½ weeks after fistulectomy. No evidence of progression of the lesions was obtained beyond 6 weeks after fistulectomy within the limits of these experiments.

The most advanced lesions were found in the area of greatest vascular development and in those animals in which the fistula had stimulated excellent collateral circulation. The most apparent physiologic explanation relates the development of the lesions to the increased blood flow through the collateral bed, the smaller arteries undergoing morphologic change as a result of the strain of excessive dilatation. The larger arteries are apparently structurally capable of adaptation to the increased volume of blood.

Whereas the basic pathologic lesions are established in the presence of the fistula and controlled by the development of the collateral circulation, these studies indicate that the lesions are progressive for a time after fistulectomy;

however, evaluation of the postfistulectomy changes is rendered somewhat difficult by the contraction of the arteries associated with regression of the collateral bed. Changes occurring during this regressive phase bear some resemblance from a pathogenetic standpoint to those observed in major visceral arteries after removal of the viscus with consequent reduction of blood flow through the arteries. They also resemble the early stages of the obliteration of the ductus arteriosus and the umbilical arteries in the infant as well as the endarteritis that occurs in arteries ligated in continuity and in the arteries of amputation stumps.[8, 9] A similar pathologic change has been observed by Vineberg[10] and Glenn and Beal[11] in studies on the fate of the internal mammary artery implanted in the myocardium. The latter authors noted the appearance as early as 3 weeks after operation of marked intimal proliferation in the partially defunctionalized but still patent artery.

SUMMARY

1. Histologic changes consisting of destruction of the internal elastic membrane and fibrous proliferation in the intima have been demonstrated in the small arteries of the collateral bed developed by femoral arteriovenous fistulas in dogs.

2. The lesions evolved during the period of the functioning fistula and progressed for a time after excision of the fistula.

3. The development of the lesions was apparently related to the strain of excessive dilatation caused by increased blood flow in the collateral vessels feeding the fistula. Progression of the lesions after fistulectomy probably resulted from the decreased blood flow coincident with contraction and regression of the collateral bed.

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ANATOMIC SKETCHES PHOTOGRAPHED ON TRANSLITE FILM

Description of a Visual Aid for Conferences and Lectures

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MOST doctors are not artists. During conferences or lectures, verbal descriptions of physical findings, pathologic lesions, or operative techniques are often supplemented by freehand blackboard sketches. All too frequently these artistic efforts are clear only to the designer. In conferences where repeated reference is made to one body location, permanent anatomic sketch outlines may be painted on a blackboard and further amplified by the use of colored chalk. However, there are definite limitations of this method for teaching and demonstration purposes.

The visual aid described herein has superior optical qualities and requires only a roentgen-ray view box and a wax pencil—accessories which are usually available in a conference room. Black on white anatomic line sketches illustrating the body region under discussion are either drawn to specification or selected from textbooks or other sources. These are photographed on to translite film* with the desired degree of enlargement. The prepared photograph is placed over the illuminated lightbox and supplemental drawing readily accomplished thereupon with a colored wax pencil.†

The photographs shown in the illustrations are examples of prepared sketches drawn to the author's specifications by the medical artist.‡ Simple line drawings in black and white, with a minimum of shading or two-tone effects, give the best optical results. We have found the 14 by 17 inch size most practical since it completely covers the standard roentgen-ray view box and renders an enlargement which is suitable for most purposes. Ordinary x-ray film may be used satisfactorily but does not produce the desirable optical qualities of translite film, which provides a background of ground glass appearance with the elimination of glare and light reflection.

The lines made in colored wax pencil are clearly visible at the back of a large conference room and form a very satisfactory contrast to the black-on-white drawing. The colored pencil lines are readily removed with a cloth or absorbent cotton pledget moistened with carbon tetrachloride, ether, benzene, or

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*Eastman Kodak Company, Rochester, N. Y.

†Dixon-Phano for glazed surfaces.

‡Drawings by Mr. Lou Barlow and photographs by Mr. Sidney Shapiro of the Medical Illustration Department of this hospital. The techniques of production of these photographs are reported elsewhere. Shapiro, S., and Barlow, L.: A New Adaption of the Black and White Transparency as a Teaching Aid, J. Biol. Photographic A. 18: 79-81, 1950.

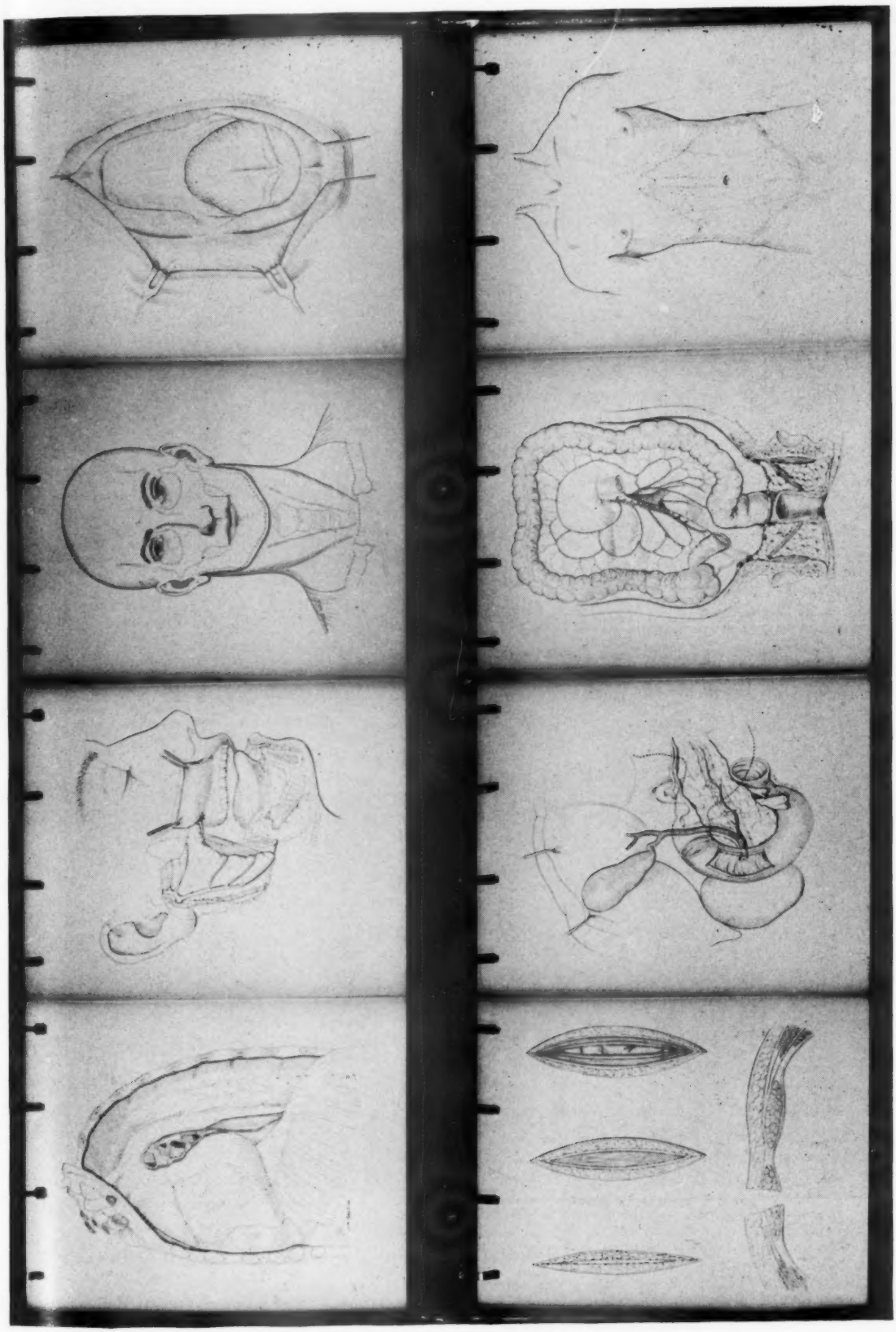
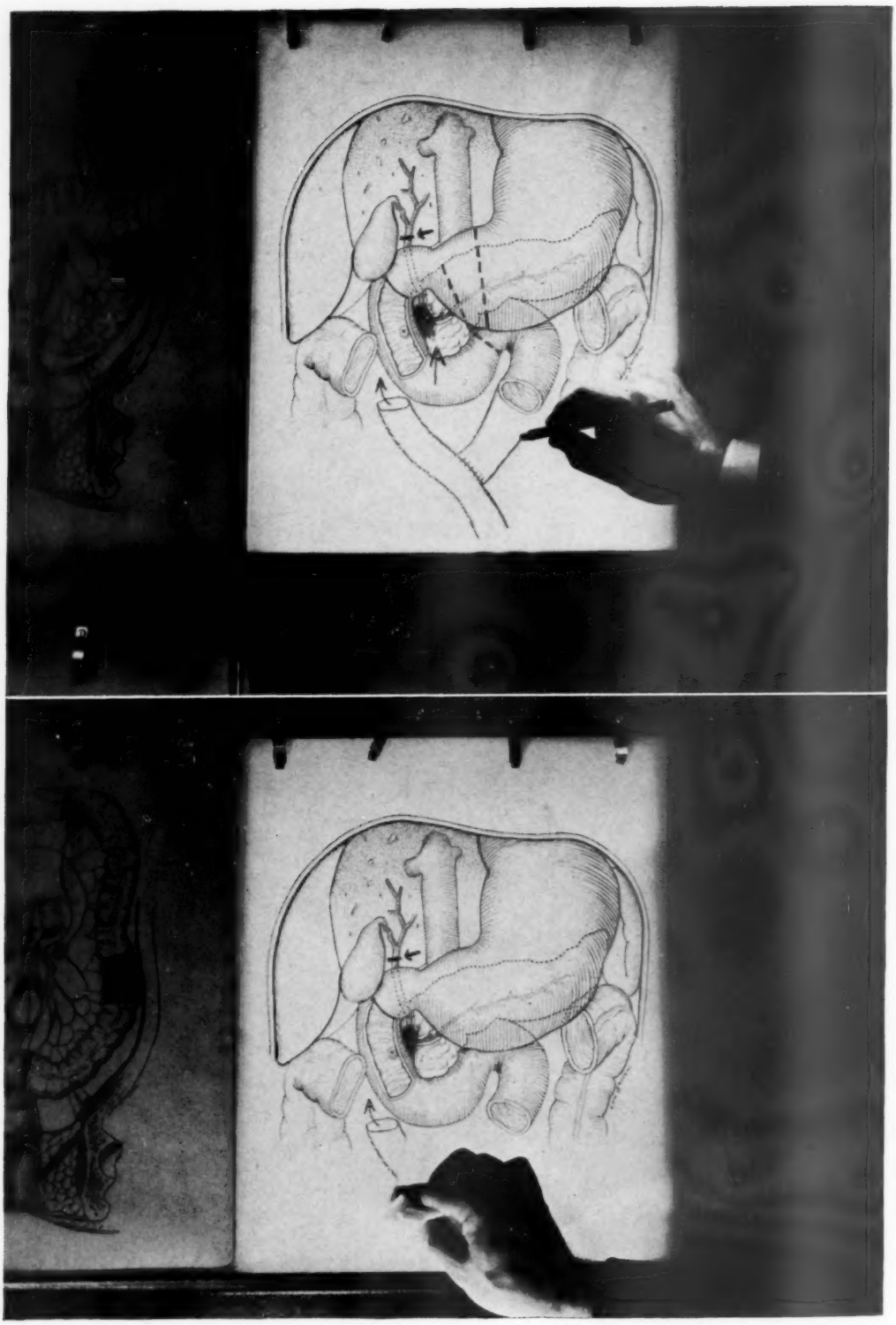


Fig. 1.—Demonstrating a variety of drawing on translite film for illustrating the location of most lesions of the head and neck. Parasagittal or lateral views may be reversed to represent right or left sides. Photographed with the transmitted light of a standard view box.

A.



B.

Fig. 2.—*A*, Demonstrating the use of waxed pencil to illustrate the location and extent of a lesion and the surgical technique used. The contrast of the colored wax pencil markings is not apparent in this black and white reproduction.

B, Same drawing after partial removal of the pencil lines with carbon tetrachchloride. Solvent completely removes wax lines without disturbing the photograph.

acetone. We have obtained best results with carbon tetrachloride, for which reason, in addition to its noninflammability, we recommend its use. The solvent has no effect on the photographed drawing.

Our experience has shown that translite film preparations have many advantages over the customary blackboard for certain purposes. The clarity and sharpness of the image must be seen to be fully appreciated. Because they supply an accurate anatomic background, many doctors have been encouraged to illustrate their descriptions of a pathologic lesion or an operative procedure. Translite films are relatively inexpensive, costing less than twice the price of standard roentgen film. They may be used repeatedly with very little wear. When needed, duplicates are readily reproduced from the original drawing. A complete series of films may be conveniently carried about in a jacket envelope. Since a view box is usually available, the films may be used on wards or small rooms where blackboard facilities are not available.

SUMMARY

The use of anatomic line drawings photographed on translite film is described as a visual aid for lectures, conferences, and demonstrations. These films are inexpensive, easily transported, and require only a roentgen-ray view box and a colored waxed pencil for demonstrations. The pencil marks are readily removed, allowing the films to be used repeatedly. Excellent optical qualities and other advantages of the method described would indicate its superiority over the conventional blackboard for certain demonstration purposes.

THE PROBLEM OF THYROID CARCINOMA

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RECOGNITION of the necessity for the removal of solitary thyroid nodules because of the high incidence of malignancy is becoming universal. No agreement has been reached, however, on the method of handling the multinodular nontoxic goiter. The validity of a regime calling for the prophylactic removal of such goiters has been questioned recently by two challenging reports. Vanderlaan[1] could find only 5 cases of carcinoma of the thyroid gland among 18,668 autopsies performed at the Boston City Hospital from 1896 to 1945. Rare neoplasms like adrenal, liver, and pituitary carcinomas were three to six times as frequently encountered as thyroid carcinoma. He concluded that carcinoma of the thyroid gland was a rare cause of death. Rogers, Asper, and Williams,[2] from the same institution, after a careful evaluation of their data, decided that prophylactic removal of all nodular goiters was not justified.

Anglem and Bradford[3] presented statistics of their own cases and discussed the reports of Vanderlaan and Rogers and associates. They concluded that, though the incidence of cancer in nontoxic multinodular goiters was not high (4.8 per cent) there were compelling reasons for removing such growths in addition to the possibiltiy of malignant degeneration. Crile and Dempsey,[4] in a review of their extensive experience, found a high incidence of malignancy in surgically removed nodular glands (10.9 per cent of nontoxic nodular goiters and 24.5 per cent of solitary nontoxic tumors). They believed, however, that the patients they encountered represented a select group chosen by internists who had already eliminated cases in which malignancy was unlikely or not suspected. Clinical judgment rather than statistical surveys served as their criterion for surgical intervention.

Despite the voluminous literature on this subject, the controversial nature of recent reports indicates the need for a more definitive approach to the problem of the nodular goiter. We have therefore felt it timely to report our experience from a large general hospital. Because most of the work has been published by surgeons, a survey made by endocrinologists commonly confronted with the necessity of making decisions on the treatment of the nodular goiter may have a certain additional value.

METHODS

The records of all patients with thyroid malignancy hospitalized in the Philadelphia General Hospital from 1936 to 1946 inclusive were analyzed. In the majority, microscopic slides from surgical or autopsy specimens were

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available and were reviewed by the present hospital pathologist.* They were then sent to six thyroid pathologists in different institutions for their diagnostic opinions.

INCIDENCE

The incidence of thyroid malignancy for the years 1936 to 1946 inclusive is outlined in Table I. The hospital files recorded 49 cases of thyroid carcinoma for this period. Fifteen were discarded after review because the diagnosis could not be established either microscopically or clinically. The 34 cases remaining were all recorded on the basis of the histologic report as proved instances of primary carcinoma of the thyroid gland. During this 11 year period 5.2 per cent of all nontoxic nodular and adenomatous goiters encountered either in surgical or in autopsy specimens were considered to be malignant. This was computed only from patients who clinically showed a nodular goiter on admission to the hospital and therefore includes only 18 of the total 34 cases of thyroid malignancy. The remaining 16 patients were not reported as having nodular goiters. Statements on the clinical charts of these 16 cases varied from "a normal thyroid" to descriptions of large masses in the neck with no resemblance to a nodular or adenomatous thyroid gland.

TABLE I. THE INCIDENCE OF THYROID MALIGNANCY AT PHILADELPHIA GENERAL HOSPITAL FROM 1936 TO 1946 INCLUSIVE

				PERCENTAGE INCIDENCE OF THYROID MALIGNANCY
Total admissions		271,803		0.013
Nodular goiter		447		4.0
Toxic	102			
Nontoxic	327			
Malignant	18	345		5.2
Thyroid malignancy		34*		
(Nodular goiter with malignancy			18)	
Total goiters (smooth and nodular)			888	3.8

*Prior to review by the group of consulting pathologists.

Slides of the tissue sections were available for study in 30 of the 34 cases considered to be primary thyroid carcinoma. Because it is well known that great difficulty may be encountered in distinguishing between thyroid hyperplasia and neoplasia[7] 7 of the leading thyroid pathologists† in the country were consulted and agreed to review these cases. The slides, together with a brief protocol of the clinical data, were sent to each pathologist in turn. In several instances one or more of the pathologists rendered a diagnosis despite what was considered an incomplete protocol or an imperfect tissue section. The 7 consultants had no knowledge of the opinions of their colleagues and accepted the material as representing cases of primary carcinoma of the thyroid gland. In Table II their diagnoses in each of the 30 cases are recorded and in Table III the percentage of agreement among them is tabulated.

*Dr. William E. Ehrich.

†Consultant pathologists were: Albert C. Broders, M.D., R. Philip Custer, M.D., William E. Ehrich, M.D., Frank W. Foote, Jr., M.D., Allen Graham, M.D., Robert C. Horn, Jr., M.D., and Tracy B. Mallory, M.D.

One of these pathologists (Dr. Graham) is a member of The Committee on Thyroid Cancer of the American Goiter Association, and two (Dr. Broders and Dr. Foote) serve as consultants.

TABLE II. DIAGNOSES OF SEVEN CONSULTANT PATHOLOGISTS (A TO G) IN THIRTY CASES OF "PRIMARY THYROID MALIGNANCY"

CASE	A	B	C	D	E	F	G
A. N.*	M	P	P	P	M	P	P
J. H.	M	M	M	M	M	M	P
M. S.	P	P	P	P	P	P	P
A. A.*	P	M	M	P	P	M	P
E. M.	M	M	M	M	M	M	M
E. P.	B	B	B	B	B	B	B
W. B.*	B	B	P	B	P	B	B ?
A. R.	P	P	P	B	P	P	P
M. A.	P	P	P	P	P	P	P
J. Y.	M	M	M	M	M	M	M
E. S.	P	P	P	P	P	P	P
A. D.	P	P	P	P	P	P	P
A. G.	P	P	P	P	P	P	P
B. F.	M	M	M	M	M	M	P
E. B.	P	P	P	P	P	P	P
L. G.	M	M	M	M	P	M	M
R. D.	P	P	M	P	P	P	P
H. B.*	P	P	M	P	P	M	P
J. H.*	P	P	M	P	P	M	P
C. F.*	M	M	M	M	P	M	P
O. J.	P	P	P	P	P	P	P
L. K.	P	P	P	P	P	P	P
M. I.	P	P	P or M	P	P	P	P
M. C.	—	P	P	P	P	P	P
V. S.*	P	P	M	M	P	M	P
E. R.*	—	P	M	P	P	P or M	M
E. S.	P	P	M	P	P	P	P
R. B.*	P	M	M	M	M	P	P
R. W.	M	M	M	M	M	M	M
T. N.*	M	P	M	M	P	P	M
Total Primary Malignancies	17	19	12	17	22	16	22

P — Primary carcinoma of the thyroid
M — Metastatic (secondary) thyroid malignancy
B — Benign
*Cases of substantial disagreement.

Examination of these data reveals that of the 30 cases of supposedly primary thyroid carcinoma, the consultant pathologists concurred with this diagnosis in only 12 to 22 instances. In one case (E. P.) all agreed upon the benign nature of the lesion. In another instance (W. B.) 4 considered the lesion benign, 2 thought it malignant and one was doubtful though he tended to agree with the majority. In a third case (A. R.) only one of the 7 consid-

TABLE III. THE PER CENT OF AGREEMENT IN DIAGNOSES OF SEVEN CONSULTANT PATHOLOGISTS (A TO G)

	A	B	C	D	E	F	G
A		86	67	86	82	79	86
B	86		71	87	83	86	77
C	67	71		72	62	82	62
D	86	87	72		77	79	77
E	82	83	62	77		79	73
F	79	86	82	79	79		72
G	86	77	62	77	73	72	
Average agreement of each pathologist with the others	81	82	69	75	76	80	75

ered the lesion benign. Thus there was substantial disagreement concerning the presence or absence of malignancy in only one case. As we are mainly concerned with the diagnosis of primary thyroid carcinoma, the differentiation of this from secondary malignant lesions in the thyroid was considered important. In Table II substantial disagreement is noted among the 7 pathologists in at least 10 cases. In 13 cases all agreed completely in their diagnoses and in an additional 6 cases all but one pathologist concurred.

In Table I the incidence of malignancy in nontoxic nodular goiter is found to be 5.2 per cent prior to a review of the histologic slides. When these figures are revised on the basis of the opinions of the consultant pathologists, a substantial decrease in incidence of primary thyroid malignancy must be made as tabulated below:

PATHOLOGIST	PER CENT
A	2.6
B	3.8
C	2.6
D	2.9
E	3.8
F	3.2
G	3.5

The average of these 7 opinions is 3.2 per cent, representing a reduction of 38 per cent in our computed incidence of primary thyroid carcinoma.

INCIDENCE FROM THE LITERATURE

In Table IV a total of 59,603 cases of thyroid disease is accumulated from 16 reports in the literature. Of these there were 1,671 thyroid malig-

TABLE IV. THE INCIDENCE OF THYROID MALIGNANCY IN THYROID DISEASE ACCUMULATED FROM SIXTEEN REPORTS IN THE LITERATURE

REFERENCE	TOTAL THYROID CASES		TOTAL MALIGNANCY		PER CENT
Bloodgood[7]		148		10	6.8
Balfour[11]		6,359		103	1.6
Speese and Brown[12]		426		22	5.2
Brenizer and McKnight[13]		5,626		84	1.5
Stout[14]		4,671		117	2.5
Hinton and Lord[6]		184		14	7.6
Cole, Slaughter, and Rossiter[15]		836		41	4.9
Cattell[16]		21,349		487	2.3
Horn, Welty, Brook, Rhoads, and Pendergrass[8]		2,079		62	2.9
Ward[9]		3,690		171	4.6
Hare[17]	Simple adenomas 1,971		236		12.0
	Multiple colloid adneomas 1,782		11		0.6
		3,753		247	6.6
Rogers, Asper, and Williams[2]		3,221		74	2.3
Hardymon and Bradshaw[18]		441		12	2.7
Frantz, Quimby, and Evans[19]		4,707		151	3.2
Anglem and Bradford[3]		1,377		46	3.3
Crile and Dempsey[4]		736		30	4.1
Totals		59,603		1,671	2.8

TABLE V. THE INCIDENCE OF THYROID MALIGNANCY IN NODULAR OR ADENOMATOUS GOITER ACCUMULATED FROM SEVEN REPORTS IN THE LITERATURE

REFERENCE	NODULAR OR ADENOMATOUS GOITER (TOXIC OR NONTOXIC)		TOTAL MALIGNANCY	PER CENT
Schlesinger, Gargill, and Saxe[20]		112	5	4.5
Hinton and Lord[6]		184	14	7.6
Cole, Slaughter, and Rossiter[15]	Nodular toxic	330	4	1.2
	Multinodular toxic	247	4	1.6
	Solitary nodular toxic	67	0	0.0
	Multinodular nontoxic	100	11	11.0
	Solitary nodular nontoxic	92	22	23.9
		836	41	4.9
Horn, Welty, Brook, Rhoads, and Pendergrass[8]		1,135	62	5.5
Ward[21]		3,690	171	4.6
Crile and Dempsey[4]		537	30	5.6
Anglem and Bradford[3]		655	44	6.7
Totals		7,149	367	5.1

nancies, an incidence of 2.8 per cent. In Table V 367 cases of thyroid malignancy were reported from 7,149 total cases of toxic and nontoxic nodular goiter, an incidence of 5.1 per cent. This may be compared with our unrevised figure of 4.0 per cent. Sufficient data for calculating the incidence of malignancy in nontoxic nodular goiters were not available, though in individual reports it is invariably greater than 5.1 per cent.

CLINICAL CHARACTERISTICS

The clinical characteristics and criteria for diagnosis have been well documented in numerous publications. It is of interest, however, to mention that in 3 of our 30 cases the diagnosis of thyroid malignancy was not suspected until surgery or post mortem, as the examining physicians had found no enlargement of the thyroid gland and no masses in the neck. A brief recapitulation of these cases follows.

E. S. (Surgical 29365, Autopsy 36592), a 45-year-old female, was admitted to the Philadelphia General Hospital with the clinical diagnosis of a lumbar spine lesion with transverse myelitis. Thyroid involvement was not suspected and thyroid enlargement was not found on examination. Exploration of the spine revealed a "pulsating tumor mass beside the bodies of the second and third lumbar vertebrae." Biopsy was performed. A bruit was heard over the mass at operation. The patient died postoperatively.

Autopsy diagnosis was nodular hyperplasia of the thyroid with a calcified focus and metastatic neoplasm of the right lung and lumbar vertebrae. Histologic examination showed primary thyroid carcinoma with metastatic thyroid carcinoma of the spinal cord.

E. B. (Autopsy 31568), a 44-year-old female, was admitted with an extramedullary spinal cord tumor opposite the first thoracic vertebral body. Duration of the history was 2 years and the patient was admitted for treatment of transverse myelopathy.

Physical examination showed no evidence of thyroid enlargement. Laminectomy was performed and a soft, necrotic, extremely vascular tumor was found in the body of the second thoracic vertebra.

Surgical pathologic diagnosis was metastatic adenocarcinoma of the thyroid, type II. The patient died postoperatively and was autopsied.

At autopsy a secondary carcinoma of the thoracic vertebrae was found. The right lateral thyroid lobe was enlarged and studded with nodules. No other sites of metastasis were discovered. The diagnosis was primary carcinoma of the thyroid gland with vertebral metastasis.

M. I. (Autopsy 39666), a 74-year-old female, was admitted with the clinical diagnosis of arteriosclerotic cardiovascular disease with heart failure and bronchopneumonia. The thyroid gland was said to be not enlarged.

X-ray diagnosis was carcinoma of the lung and bronchopneumonia.

Post-mortem examination revealed thyroid malignancy with metastasis to the lung, liver, and myocardium.

DISCUSSION

In attempting to compute the incidence of malignancy encountered among nontoxic multinodular goiters in a large general hospital, the original impression that 5.2 per cent of these goiters were malignant required revision upon further study. When the consultative services of a group of pathologists whose opinions in thyroid histology are authoritative were utilized, almost one-half of our supposedly pathologically proved cases of primary thyroid carcinoma had to be discarded, and the incidence of primary malignancy in nodular goiters was reduced from 5.2 to 3.2 per cent, a reduction in incidence of 38 per cent from the original estimate. Important differences of opinion were evident even among the 7 consultants. Consultants A, B, and F, including our own pathologist, who found the least area of disagreement, still concurred with their colleagues in only 81, 82, and 80 per cent of the 30 cases, respectively. These patients were hospital subjects, many of whom were admitted in a terminal state, so that absolute percentage figures cannot be compared with all of the other studies reported in the literature; yet our unrevised incidence of 4.0 per cent malignancy among our toxic and nontoxic nodular goiters is not greatly different from the 5.1 per cent figure accumulated from the literature. One wonders whether a similar review by several pathologists of the cases of other series would produce a substantial revision of the incidence of primary thyroid malignancy. The marked discrepancy in the statistics from various series (from 4.6 per cent in Ward's series to 11 per cent in Cole's) suggests that such might well be the case. Again, in Table VI the findings at the Philadelphia General Hospital are added to a table prepared by Rogers, Asper, and Williams.[2] The percentage incidence of thyroid malignancy encountered at Philadelphia General Hospital, Johns Hopkins, and Massachusetts General Hospital is respectively 3, 9, and 10 times greater than that reported at Boston City Hospital. It is difficult to explain these marked differences. While it is possible that Johns Hopkins and Massachusetts General Hospital attract more cases of thyroid disease, differences in pathologic criteria for making the diagnosis may be an important factor. A similar review of the cases from these hospitals might result in a similar striking reduction in the incidence of primary thyroid carcinoma.

The question of diagnosis of thyroid malignancy becomes further complicated by the fact that a certain number of cases develop from foci so small that the thyroid may appear normal to the palpating finger (so-called car-

TABLE VI. THE INCIDENCE OF THYROID MALIGNANCY AMONG TOTAL ADMISSIONS TO FOUR DIFFERENT HOSPITALS

INCIDENCE OF MALIGNANCY IN TOTAL ADMISSIONS	HOSPITAL	ADMISSION	ALL GOITERS	TOXIC GOITERS			NONTOXIC GOITERS			MALIGNANCY	
				UNSPEC. TYPE	NOD.	DIFF.	UNSPEC. TYPE	NOD.	DIFF.		
0.0042	Boston City 1931-1942	406,402	1,125		125	306	16	551	127	10 7*	17
0.013	Philadelphia General 1936-1946	271,803	888		102	180		345	146	34†	34
0.039	Johns Hopkins 1940-1944	77,399	717	24	72	210	34	161	216		30
0.044	Massachusetts General 1937-1944	61,117	1,379		180	391		751	57	24 3*	27

*Not confirmed by histologic examination but clinical course compatible with diagnosis.
†Prior to review by consulting pathologists.

cinoma in situ) and the diagnosis may be established only after metastasis occurs. The thyroid was said to be normal or not enlarged on examination in 3 of the cases in this series. If 3 of 30 cases were not even suspected clinically and only 3.2 per cent of all palpably nodular goiters were malignant, are we justified in recommending the wholesale removal of all multinodular goiters? It might also be mentioned here that a palpably nodular goiter was described in several instances where our cooperating pathologists disagreed as to whether the malignancy was primary or secondary in the thyroid. Certainly one would not recommend thyroidectomy for a metastatic thyroid nodule from some distant primary source.

A review of the literature fails to offer a satisfactory solution to this problem. The incidence of multinodular malignant goiters varies from 4.5 per cent (Schlesinger and associates[20]) to 11 per cent (Cole and co-workers[15]). Opinions vary from those who advocate wholesale removal of all nontoxic nodular goiters[6-9] to those who believe that such prophylactic surgery is completely unwarranted.[2, 4] Hinton and Lord[6] felt that the incidence of clinically unsuspected cancer in thyroid nodules is higher than in breast nodules, whereas Crile and Dempsey[4] stated, "In the goitrous thyroid, carcinoma does not develop as frequently as in the apparently normal breast."

The true incidence of malignancy in nontoxic nodular and adenomatous goiters is not known at the present time. This study demonstrates that diagnoses based on tissue sections may be expected to vary appreciably among qualified pathologists. Until ample and accurate data are available and pathologic criteria for diagnosis are more firmly established, dogmatic statements concerning the proper therapeutic approach to the multinodular nontoxic goiter carry no authority. The Committee on Thyroid Cancer of the American Goiter Association was established in 1940 for the purpose of collecting clinical and pathologic data from all such cases throughout the country. To date, fewer than 500 have been submitted to the Committee.[10] This small number undoubtedly has not resulted from apathy on the part of the medical profession but more likely from ignorance of the existence and purposes of the Thyroid Cancer Committee, a situation which should be rectified by a wide publicity program disseminated throughout the medical centers of the country. Only until this Committee has had the opportunity to analyze very large numbers of cases of nodular goiter and thyroid malignancy can decisive criteria be established to aid in the future therapy of such patients. Until that time the authors must agree with Crile and Dempsey, who concluded their critical review by suggesting that indications for removal of nodular goiters be based on clinical judgment rather than on statistical surveys.

SUMMARY

A group of 7 eminent consultant pathologists have reviewed the tissue sections of a series of cases of supposedly primary thyroid carcinoma. The originally computed incidence of primary malignancy of 5.2 per cent required revision to 3.2 per cent, a reduction of 38 per cent. The consultants agreed

that only 12 to 22 of the 30 cases were instances of primary thyroid malignancy; the remainder were secondary (metastatic) malignancy, and 2 or 3 cases were benign lesions. Agreement among the pathologists varied from 69 to 82 per cent. The true incidence of thyroid malignancy in nontoxic nodular and adenomatous goiters is unknown at the present time and pathologic criteria are not yet firmly established.

ADDENDUM

At the completion of this study, Tables II and III were sent to the 7 consulting pathologists for their examination and criticism. It was decided to add any opinions received to the manuscript. The only one who requested that his further opinions be added was pathologist G., who wrote, "I am confident that, if diagnoses made with reservation are taken into consideration and genuinely doubtful cases excluded, the consultant pathologists participating in this study would reach very substantial agreement as to what is and what is not carcinoma of the thyroid gland. It is further my belief that the individual experienced pathologist, carefully studying his own material in collaboration with his clinical colleagues, has the best opportunity to evaluate the problem cases and to contribute to knowledge of thyroid carcinoma."

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Case Reports

A CASE REPORT OF LEIOMYOSARCOMA OF THE DUODENUM

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THE first case of leiomyosarcoma of the duodenum was reported by von Salis,[10] in 1920. The fact that less than fifteen cases have been reported in the literature[5-7, 9, 10] to date justifies the presentation of the following case.

CASE REPORT

J. A., a 56-year-old white man, a foundry worker, was admitted to the Dayton Veterans Administration Hospital on Jan. 10, 1950, complaining of progressive weakness, lethargy, and loss of weight for the past five months. The patient had been in good health prior to August of 1949, when he began experiencing nocturnal chills associated with fever, "flushing of the face," and profuse diaphoresis. These episodes occurred at intervals of two to three times a week and were not accompanied by nausea, vomiting, nor abdominal distress of any kind. Shortly after the onset of these symptoms, the patient began to feel weak and lethargic, noting progressive weight loss amounting to 54 pounds in a period of eight months. He stated that he had been constipated for a number of years and had resorted to the frequent use of laxatives. Approximately three and one-half months prior to admission the patient noted the onset of progressive painless jaundice associated with light stools and dark urine. There was no history of tarry or bloody stools, and no change in bowel habits.

Past history was noncontributory, including a negative familial history of cancer. The patient denied excessive use of alcohol and gave no history of exposure to toxic chemicals.

Physical examination upon admission revealed a middle-aged, chronically ill white man in no acute pain, who evidenced some recent weight loss. The patient appeared somewhat lethargic, with a temperature of 99°, pulse 84, respiration 16, blood pressure 118/80. The skin and sclerae showed a definite icteric coloration. There was no lymphadenopathy. The chest was clear to percussion and auscultation, and the cardiovascular system was normal. The abdomen was scaphoid, soft, and nontender, with an ancient healed right lower quadrant appendectomy scar. The liver was two to three fingerbreadths below the right costal margin, the edge was smooth and nontender, and the gall bladder was not palpable. There was no evidence of an intra-abdominal mass nor ascites. The remainder of the physical findings, including rectal examination, were negative.

Laboratory Data.—(Laboratory tests were performed upon admission unless otherwise specified.) Blood tests were: red blood cell count 3.3 million; hemoglobin 10 Gm.; white blood cell count 12,500; neutrophils 78 per cent; lymphocytes 18 per cent; monocytes 4. Urine tests were: color, dark amber; specific gravity 1.010; albumin negative; sugar negative; microscopic examination negative blood urea nitrogen 17. Serology was negative. Liver and biliary function tests were: prothrombin time, 90, 64, and 100 per cent (three determinations between Jan. 13 and Jan. 27, 1950; vitamin K was administered after Jan.

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13, 1950); icterus index 25 (on admission), 28 (Jan. 13, 1950), 42 (Jan. 27, 1950); van den Bergh test 1.5 direct, 1.2 indirect (on admission); 1 direct, 1.4 indirect (Jan. 20, 1950); total bilirubin 2.7; cephalin flocculation negative (on admission), 2+ (Jan. 27, 1950); thymol turbidity 1.5; alkaline phosphatase 18.4; cholesterol 189; cholesterol ester ratio 86 per cent; bromsulfalein dye retention 100 per cent; serum amylase 225, 250, 372, 516 (determinations between Jan. 10, 1950, and Jan. 27, 1950, normal laboratory value less than 200); urine urobilinogen .23 for 192 c.c. Gastric analysis was: total acidity 44, 60, 66. Free acid was 9, 7, 30 (fifteen-minute aspirated samples). Stools were light yellow in color with no abnormal fat, no blood, and no parasites. Electrocardiogram was normal.

Fig. 1.—A linear pressure defect is present across the distal portion of the duodenal cap. There appears to be a mass protruding into the lumen from inferior portion of the second part of the duodenum.

Roentgenographic Findings.—The chest was negative. The gall bladder was not visualized and no stones were demonstrated. Barium enema was negative except for the presence of two small incidental sigmoidal diverticula. Gastrointestinal series showed the esophagus and stomach were relatively normal. There was no organic lesion of the duodenum present, though there appeared to be a linear pressure defect across the distal cap. The duodenal loop was found to be abnormal. There appeared to be a mass protruding into the duodenum in the inferior portion of the descending segment of the loop (Fig. 1). This was slightly more distal than in the case of an ampullary malignancy, hence it was the opinion of the roentgenologist that these changes were secondary to a malignancy of the head of the pancreas. The stomach emptied normally at the end of six hours.

Progress in the Hospital.—The patient's temperature ranged between 99.4° and 102.4° at intervals of two to four days, each painles febrile episode being associated with diaphoresis and some flushing of the skin of the face. He was given iron, vitamin K, 9 units of

blood, and supportive liver medication with some improvement in his general physical condition in regard to weakness and lethargy. The jaundice, however, continued to increase, and the liver profile showed progression of hepatic insufficiency. A preoperative diagnosis of carcinoma of the head of the pancreas was made, based largely upon the x-ray findings accompanied by progressive painless jaundice.

Operation was performed on Jan. 30, 1950. The abdomen was opened through a transverse incision. There was a rather deep jaundice of the bowel and intra-abdominal structures. The liver was moderately enlarged, its surface was smooth, and there was no evidence of carcinomatous implants. The gall bladder was enlarged two and one-half times its normal size, and the common duct was 2½ cm. in diameter. The stomach and spleen were normal. On further exploration there was found a hard rounded mass approximately the size of a golf ball, which seemed to involve the head of the pancreas. A few small glands were found near the duodenal terminus of the common duct, one of which was removed for a frozen section and was reported negative. There was no evidence of fixation to surrounding structures, consequently the tumor was resected. This involved the performance of a subtotal pancreatectomy, complete duodenectomy, and a choledochopancreatico- and gastrojejunostomy.

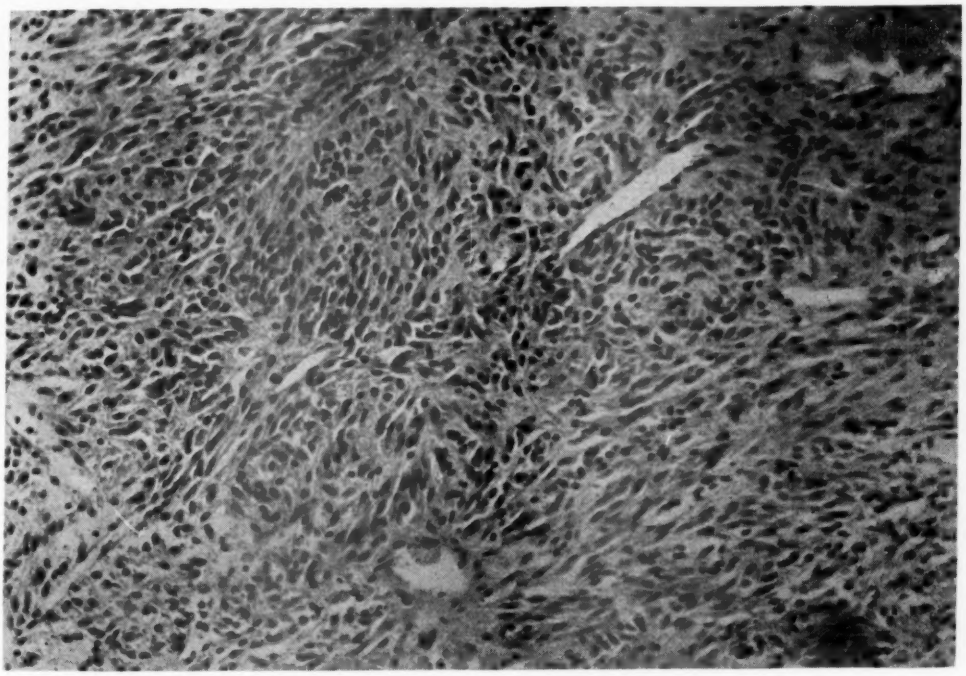

Fig. 2.—Section (×43) reveals highly cellular tissue arranged in whorls and strands. The nuclei are large, hyperchromatic, and contain many mitotic figures. There is abundant light-staining cytoplasm, moderate pleomorphism, little evidence of vascularity, and no blood vessel invasion.

Pathologic Report.—

Gross: The specimen consisted of the duodenum and head of the pancreas. A small, firm, rubbery, fibrous tumor, approximately 2.5 cm. in diameter, was located within the wall of the duodenum, bulging into its lumen from the posterior medial aspect of its second portion immediately below the ampulla of Vater. The overlying mucosa was intact. The tumor mass appeared encapsulated from the surrounding duodenal wall, pancreas, and duct system. The common duct and pancreatic duct emptied separately into the ampulla of Vater, their entrances being compressed from beneath by the tumor. There was no gross evidence of invasion of either duct.

Microscopic: The section was composed of an encapsulated mass of highly cellular tissue arranged in whorls and strands and interlacing in all directions (Fig. 2). There was a uniform preponderance of large nuclei throughout the section. These nuclei were large, hyperchromatic, and contained many mitotic figures. There was abundant collagenous cytoplasm, moderate pleomorphism, little evidence of vascularity, and no blood vessel invasion. A lymph node from the vicinity of the common duct showed no evidence of tumor. The adjoining pancreas showed no invasion by tumor but there were a diffuse interlobular fibrosis, fatty infiltration, and invasion by chronic inflammatory cells of the interstitial spaces. Pathologic diagnosis was: (1) leiomyosarcoma of the periampullary portion of the duodenum, and (2) chronic interstitial pancreatitis.

The postoperative course was complicated by pancreatic and jejunal fistulas, both of which closed spontaneously. The patient was discharged asymptomatic three months after operation.

Tumors of the small intestine are rare clinical entities frequently producing gastrointestinal symptoms which are often interpreted as of functional origin. Unless one is cognizant of the possible significance of these symptoms, valuable time may be lost prior to establishing the correct diagnosis.

Raiford[8] reported that benign small intestinal tumors account for 23.8 per cent of all benign neoplasms of the gastrointestinal tract, nearly all of these occurring as intraluminal growths. These tumors are usually silent and discovered at autopsy; however, because of the location of the growth in relation to the bowel, they may cause intussusception and intestinal obstruction. While the incidence of malignant degeneration of benign tumors has not been definitely established, it is most often reported between 7 to 10 per cent.[1, 4, 8] The benign group includes adenomas, leiomyomas, fibromas, lipomas, hemangiomas, pancreatic rests, and neurogenic tumors.[1]

Adenomas comprise nearly one-third of all benign tumors of the small intestine. These small neoplasms are predominately papillomatous or polypoid in form and are most frequently found in the ileum and duodenum as single or multiple tumors. Multiple adenomatosis may be familial. These tumors seldom become sufficiently large to produce intestinal obstruction.[3]

Fibromas also occur most frequently in the ileum and rarely in the duodenum, and, although their size may vary from that of a split pea to over eight inches in diameter, they rarely cause symptoms since the majority are extraluminal. On rare occasions the intraluminal variety is propulsed by peristalsis along the distal bowel, invaginating the attached segment of intestine, and producing intussusception.

Lipomas have a predilection for the ileum but are occasionally found in the jejunum and rarely in the duodenum. These tumors, like adenomas, may be single or multiple and upon occasion occur as a manifestation of diffuse lipomatosis. Although they are usually silent intraluminal growths, rarely becoming detached and passing through the intestinal tract, they may produce intestinal obstruction by intussusception, or cause gastrointestinal hemorrhage.

Hemangiomas of the small intestine, usually multiple and intraluminal, vary in size and degree of vascularity from pin-point varicosities to large ulcerating cavernous lesions occurring most frequently in the jejunum and ileum, often giving rise to profound hemorrhages.

Pancreatic rests, arising from embryonic pancreatic anlages,[1] are most often encountered in the duodenum. These tumors, composed of pancreatic tissue, are usually asymptomatic, rarely producing obstruction or bleeding. The incidence of malignant degeneration is a matter of question and has not yet been determined.

Neurogenic tumors are among the rarest of benign tumors of the small intestine and most commonly are small asymptomatic lesions, discovered at routine autopsy.

Various types of cysts, including the so-called gas cysts, endometriomas, tumors of reticuloendothelial cells, and hamartomas have been reported as occurring in the small intestine.[4] These are pathologic rarities beyond the scope of this presentation.

Leiomyomas are second in frequency to adenomas, and, with the exception of the stomach, these tumors occur more frequently in the small intestine than in any other portion of the gastrointestinal tract. The ileum is the site of predilection in the small bowel, the tumors being either extra- or intraluminal, the latter more often producing intestinal obstruction. Leiomyomas often cause considerable hemorrhage into the intestinal tract because of their predisposition to undergo central necrosis. It has been estimated that 10 to 20 per cent[1] of leiomyomas undergo sarcomatous degeneration. This represents almost twice the malignant propensity of the other benign small bowel tumors.

Malignant tumors of the small intestine are reported by various authorities[1, 2, 4, 8] as ranging in incidence from 3 to 6 per cent of all gastrointestinal malignancies. Raiford reported that of 776 malignant gastrointestinal tract tumors which he reviewed in 1938, only 28, or 4.9 per cent, occurred in the small intestine.

The most common malignant tumor of the small intestine is carcinoma, usually located in the second portion of the duodenum, and often clinically indistinguishable from carcinoma of the extrahepatic bile ducts, common duct, and head of the pancreas. The symptoms produced by carcinoma of the duodenum are directly dependent upon the location of the tumor.

The case presented demonstrates a malignant tumor, probably of benign origin, in such a location that the symptoms and signs prior to operation rendered it indistinguishable from the more frequently occurring malignant tumors found in this region.

Leiomyosarcoma, as well as the benign tumors, should be considered in the differential diagnosis of all lesions producing painless obstructive jaundice.

SUMMARY

1. A case of a periampullary leiomyosarcoma producing progressive painless jaundice, weight loss, lethargy, fever, and anemia has been presented.

2. These tumors offer a much better prognosis than the more common carcinomas of the pancreatic head and bile duct system and should be considered in the differential diagnosis of lesions in this region.

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TREATMENT OF TUBERCULOSIS OF THE STERNUM BY LOCAL EXCISION

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IN SPITE of the fact that it contains a considerable amount of cancellous bone and is subcutaneous in position, the sternum is rarely the site of local disease. This is especially true of tuberculosis. Wassersug[1] found only twelve patients with disease of the sternum among 1,134 patients with bone and joint tuberculosis (an incidence of 1.1 per cent) which were treated at the Lakeview Sanatorium. If one includes disease of the sternoclavicular joint and of the costal cartilages, the number is considerably increased.

The disease is more common in young adults, but may occur at any age. It may be present as a solitary focus, but usually it is secondary to disease elsewhere, especially pulmonary or pleural tuberculosis. It is probable that in the majority of the cases the sternum is involved by extension of the disease from glands in the anterior mediastinum.

The onset is insidious and usually the first symptom is a soft swelling over the sternum. This is slightly tender and as it increases in size it becomes fluctuant and approaches the skin. If aspirated it will be found to contain rather thick white or gray pus, and acid-fast bacilli may be demonstrated in the smears by culture or by guinea pig innoculation. The margins of the swelling are indurated and in the center a craterlike depression is palpable; the bottom of this may be formed by a depression in the sternum.

Unless the natural course of the disease is interrupted by treatment, the cold abscess ruptures and forms one or more chronic sinuses which may drain for months or years and eventually heal permanently, or heal and reopen or be followed by another cold abscess. The amount of destruction of the sternum may be slight or considerable and the bone may be perforated in one or more places by the disease, or the erosion may indent or even sever the body of the bone and destroy a large part of it. New bone production is minimal and sequestra are rarely seen. The destruction of the bone may be seen in an anteroposterior sectional radiograph (laminogram) (Rudenholz[2]) or in a lateral view of the sternum.

The patient may run a low-grade fever, but the general condition is usually good unless there is deterioration from disease elsewhere. Tuberculosis of the sternum must be differentiated from pyogenic or other infections and neoplasms involving this bone.

We know of no standard treatment of the condition. Wassersug stated that the patients reported by him were treated by incision and curettage, incision and drainage, aspiration and resection of the sinuses, simple aspiration,

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or no treatment. Sinuses formed in all. In five of the patients the sinuses healed after months or years while under treatment in the hospital. Three of the patients died of tuberculosis elsewhere, one was lost sight of (probably dead), and in four the sinuses were still draining at the time of the report. He recommended conservative treatment and thought that surgery was not necessary.

The only other paper on the subject which we could find in recent literature is one by Herrick,[3] who reported a single patient in whom the abscess was aspirated three times and was treated daily for three months with ultraviolet light. The patient was in the Veterans Hospital for six months or more and was discharged with the lesion apparently healed. The diagnosis in this case was proved by guinea pig innoculation with pus from the abscess. The case reported by Johnson[4] was obviously a severe pyogenic infection and not tuberculosis.

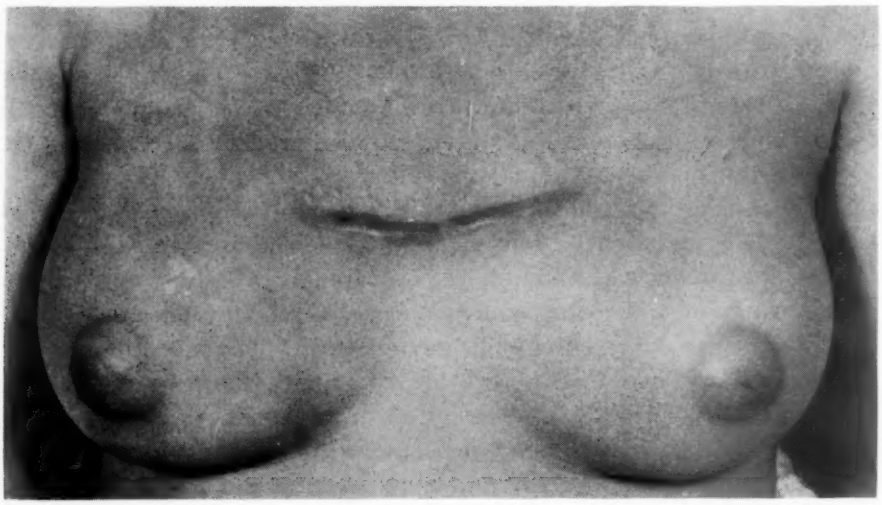

Fig. 1.—Anterior view of the first patient (Case 1) after operation.

Recently we have had two cases of tuberculosis of the sternum under our care. Based upon our experience with other forms of bone tuberculosis, it seemed to us that resection of the diseased bone offered the best chance of a cure.

This opinion was strengthened by the experience of one of us (J.A.K.) with two cases of tuberculosis of the sternoclavicular joint and three cases of tuberculosis of the pubic synchondrosis. In each of these the diseased area was resected and the wound sutured, and apparently the disease was cured permanently. Consequently our two patients with tuberculosis of the sternum were operated upon. The case reports are as follows.

Case 1.—C. F. S. (151266), a 16-year-old white girl, first noted the appearance of a small tender mass over the midsternal region in July, 1947. Because this mass continued to enlarge gradually and to remain tender, the patient presented herself for treatment on Aug. 16, 1947. At this time a tender fluctuant mass, 3 cm. across, was noted over the body of the sternum.

It was aspirated and a small amount of thick pus was obtained. Acid-fast bacilli were demonstrated in the pus; however, the innoculated guinea pig died prematurely and no gross evidence of tuberculosis was present at autopsy. Roentgenograms of the chest and sternum were taken and reported as negative.

The patient was admitted to the Orthopedic Surgery Service at Barnes Hospital on Sept. 22, 1947. She was entirely asymptomatic except for the persistent and gradually enlarging presternal mass. Its size was now approximately 4 cm. in diameter and it was elevated 1.5 cm. There was definite fluctuation and moderate tenderness. The lesion was located over the body of the sternum just inferior to the sternomanubrial joint. Roentgenograms of the chest were again reported as negative. Routine laboratory examinations were entirely normal.

On Sept. 25, 1947, the abscess was aspirated and 10 c.c. of thick, creamy, yellow-tan material was obtained. Two grams of streptomycin were injected into the abscess after the aspiration. On Sept. 30, 1947, this procedure was repeated. Acid-fast bacilli were demonstrable in the aspirated material on both occasions by staining and guinea pig innoculation.

After the patient had been on 2 Gm. of streptomycin per day for three weeks, a subtotal resection of the sternum was performed on Oct. 16, 1947. A transverse incision was made and the abscess was mobilized completely except at its attachment to the sternum. The first portion of the body of the sternum was then freed from the manubrium above, the costal cartilages laterally, and the second portion of the sternal body below by an osteotome and sharp dissection. The entire first portion of the sternal body along with the abscess was removed in toto, and care was exerted to preserve the periosteum on its dorsal surface without entering the mediastinum. The wound was closed tightly in layers after irrigation with sulfonamide solution and topical implantation of 1 Gm. of streptomycin. A pressure dressing was applied.

On gross examination, the abscess was found to be thick walled and filled with caseous material. Two sinuses were present in the upper portion of the sternal body, one on each side, but not involving the costal cartilages. Microscopic sections of the abscess and bone showed classic tuberculosis with tubercles and caseous necrosis.

The patient's postoperative course was uneventful and the operative wound healed primarily. Streptomycin was discontinued on the fourteenth postoperative day and the patient was discharged on the twenty-third postoperative day.

She was seen at regular intervals in the Orthopedic Surgery Outpatient Clinic for one year. During this time the patient was asymptomatic and there was no evidence of further infection in the sternum or any other part of the body. There was no disability or discomfort or deformity at the site of the resected portion of the sternum and the bone appeared to have regenerated in this area. Chest films were repeatedly negative. Early in 1949 this patient moved from St. Louis and is now lost for follow-up.

Case 2.—A. B. (173,210), a 40-year-old Negro man, was first admitted to Barnes Hospital on June 15, 1949, because of gradually increasing weakness, weight loss, night sweats, and fever of approximately one year's duration. On physical examination the liver edge was down 3 cm. and the spleen 6 cm. No peripheral lymph nodes of pathologic significance were palpable. The remainder of the physical findings were essentially normal. During the hospital course, the patient ran an irregular fever, spiking daily to around 39° C. A single blood culture was positive for organisms thought to be brucella; agglutinations for brucellosis, however, were negative. A sternal marrow puncture was performed on June 18, 1949, and was considered within normal limits. On June 23, 1949, an axillary lymph node that had appeared while in the hospital was biopsied and showed only hyperplasia microscopically. Because of the persistence of the hepatomegaly and splenomegaly, an exploratory laparotomy and liver biopsy were performed on June 29, 1949. The sections of liver showed a granulomatous lesion of undetermined etiology. Brucellosis and tuberculosis were both considered by the pathologist, but acid-fast stains failed to reveal tubercle bacilli. The patient was given aureomycin as a therapeutic test for brucellosis, but the fever was not lowered by this regime. He also received two weeks of intensive penicillin therapy because of central nervous system syphilis. A chest roentgenogram previously

considered negative was repeated and showed a destructive lesion of the right ninth rib in the mid-axillary line. The right ninth rib was biopsied and caseopurulent material was obtained. Microscopically this lesion was compatible with tuberculosis and seven acid-fast bacilli were identified in the material. Dihydrostreptomycin (2.0 Gm. per day) was given for 42 days. The rib biopsy incision healed per primam and there was gradual defervescence. The patient was discharged on Sept. 23, 1949, to be followed in the Out-patient Clinic.

On Dec. 1, 1949, the patient first noticed a painless swelling over the mid-sternum. This mass gradually increased in size until Dec. 20, 1949, when he was readmitted to Barnes Hospital. At this time the lesion measured 3 cm. in diameter; it was soft, fluctuant, non-tender, and was not hot. It was located at the fifth chondrosternal junction on the right side. The liver and spleen were again palpably enlarged, but the chest was entirely negative to physical examination. Laboratory findings were within normal limits. The pre-sternal abscess was aspirated on several occasions and the material obtained was smeared, cultured, and injected into guinea pigs. Acid-fast bacilli were recovered from two guinea

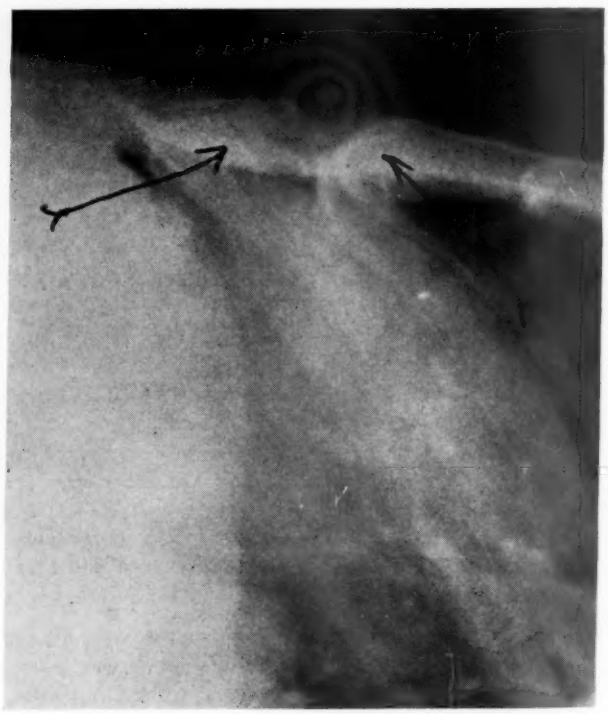

Fig. 2.—Lateral view of sternum of the second patient (Case 2) before operation. Note rarefaction at junction of middle and lower segments.

pigs, but the smears and cultures were negative. The patient was placed on 1.0 Gm. of dihydrostreptomycin per day and 10 Gm. of para-aminosalicylic acid per day. Roentgenograms and laminograms of the sternum revealed a destructive process of the inferior body and xiphoid process, as well as the chondrosternal junction on the left.

The inferior one-third of the sternum was resected on Jan. 13, 1950. The skin overlying the abscess was widely excised and the abscess wall was carefully preserved intact. By dividing the costal cartilages laterally and the mid-portion of the sternum superiorly with an osteotome, it was possible to remove the involved bone and abscess en bloc. However, there was complete erosion through the sternum and a large abscess found in the anterior mediastinum was limited by a very dense fibrous membrane from the underlying thoracic structures. This abscess was completely evacuated and its wall was carefully

curetted. Three grams of streptomycin were placed in the operative field and the wound was closed primarily after superior and inferior relaxing skin incisions had been made. Split-thickness skin grafts were necessary to fill in the defects caused by the relaxing incisions. A pressure dressing was applied.

Postoperatively the patient was put on 1 Gm. of streptomycin per day in addition to routine penicillin therapy. The skin grafts in the relaxing incisions healed satisfactorily, but the primary wound edges separated and became necrotic. The resultant defect was wet dressed and allowed to granulate in until the thirtieth postoperative day, when a split-thickness skin graft was used to complete the closure. This graft was entirely successful. The patient was then discharged from the hospital on Feb. 24, 1950. He has been followed in the Outpatient Clinic since that time. He is now gaining weight and is afebrile. There is no evidence of drainage or activity about the scar over the sternum. This defect has decreased considerably in size and its base is formed by dense scar tissue covered by the skin graft. He does not have enough trouble to warrant excision and suture of the grafted area. The disease is still active in the rib after treatment with streptomycin, and there is a small sinus draining from this area. We believe that this lesion also should be excised.

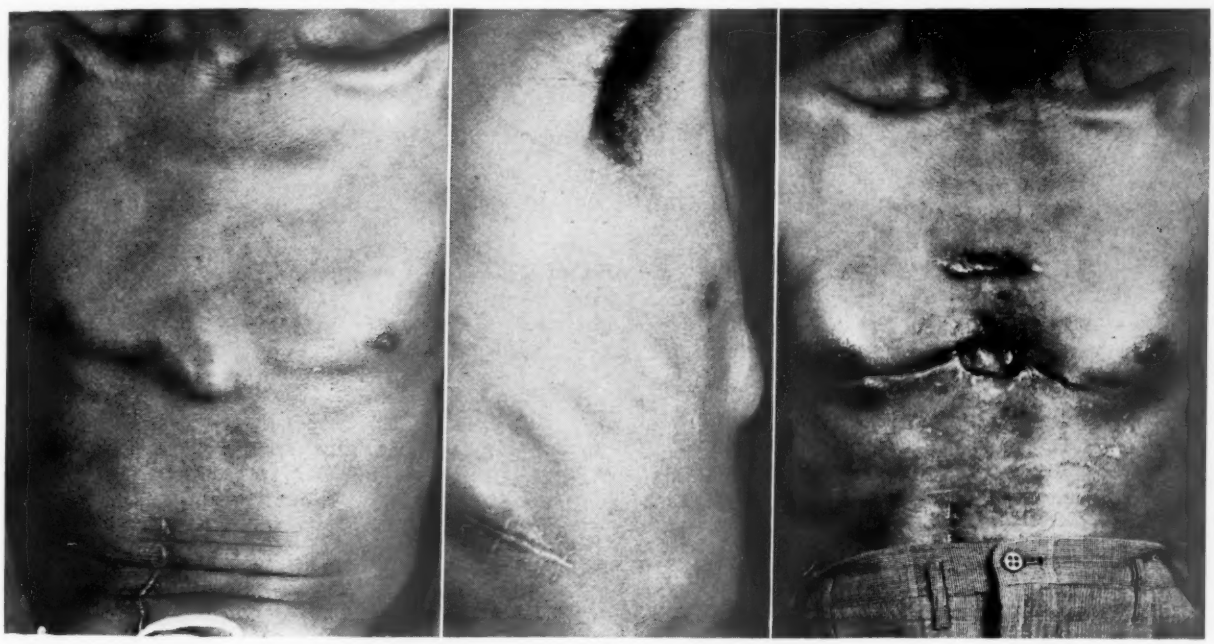

Fig. 3.—Anterior and lateral views of the second patient (Case 2) before operation and an anterior view about five months after operation. The scar from the rib biopsy is visible in the lateral view.

DISCUSSION

While it is quite possible that eventually the disease would have healed in the aforementioned patients, it is probable that this would have been a matter of months in the first patient (Case 1), and of years in the second (Case 2), and it is possible that the disease might have spread elsewhere or recurred. We believe that the local disease is now permanently cured in each case.

In the first patient the operation was done early in the disease, cure was prompt, and the convalescence uneventful. In the second patient the disease was more advanced and involved the skin, which was thin and shiny over the

apex of the swelling; rupture of the abscess was imminent. Consequently, a rather wide area of skin was excised with the abscess and lower portion of the sternum and closure was difficult. Also there was a large dead space directly beneath the skin and the base of this was diseased; consequently, the skin margins separated and the defect was covered with a split skin graft. It would probably have been better to have let the abscess rupture and the sinus drain for a few months and to operate when the disease was less active.

In removing the sternum the bone was cut transversely with a thin sharp osteotome, a V-shaped trench being cut across the superficial surface. This was deepened gradually until the posterior surface was approached, then the loose piece was pried up with a periosteal elevator. In this way there is very little danger of injuring the underlying structures. The costal cartilages were then cut with bone forceps and the involved piece of the sternum with the overlying abscess was removed in one piece. The cartilages were excised back to what appeared to be normal tissue, but no effort was made to remove the entire cartilage or to cover the end with perichondrium as advocated by Maier.[5] Likewise, no effort was made to repair the defect by a graft or a prothesis such as the tantalum plate used by Griswold.[6] In our two patients repair of the defect has not been necessary as it has not caused any disability, and the same was true of the cases in which the sternoclavicular joint or the pubis was excised. However, the defect in Griswold's case was larger than it was in either of ours.

CONCLUSION

Local excision of the diseased area is a safe and relatively sure method of effecting a prompt cure of tuberculosis of the sternum.

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HAMARTOMA OF THE LIVER

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INTRODUCTION

BENIGN tumors of the liver have always been considered a rarity. Bockus[1] stated that they may occur at any age but the majority are found in adults of middle age. In the past, benign hemangiomas have accounted for the majority of nonmalignant hepatic tumors.

In a recent publication, Kay and Talbert[2] added two cases of hamartoma or benign adenoma to the literature and reviewed seven similar cases.

CASE PROTOCOL

K. H. was first seen at the age of 1 year and 10 months, when she was brought to the clinic because of an abdominal mass in the right side of the abdomen. The infant was a female, born at full-term delivery, and was normal in every other way. Her mother stated the child's abdomen had always seemed a little enlarged. There was no history of vomiting or diarrhea or change in bowel habit.

Upon examination she was found to weigh 26 pounds. The heart and lung findings were negative, and a large, firm mass filling the entire right abdomen semed continuous with the liver above. The extremities and reflexes were negative. There was no edema and no sensation of ascitic fluid in the abdomen.

Laboratory examination revealed a hemoglobin of 8.5 Gm. with a red cell count of 3,120,000 and a white cell count of 7,800. Urine was normal.

Scout film of the chest was negative. A film of the abdomen revealed a soft tissue mass in the right kidney area. An intravenous pyelogram revealed that the mass did not arise from the apparently normal right kidney. At the conclusion of the examination, the differential diagnosis was: renal tumor, cystic in nature, possibly a Wilms,' or hepatomegaly, either cystic or tumorous.

On Feb. 14, 1950, the child was operated upon, and a large, solitary tumor arising from the right lobe of the liver was found. A partial hepatectomy was done.

The specimen was found by the pathologist to consist of a tumor mass measuring 15 by 16 by 8 cm. and having a total weight of 790 Gm. This tumor mass was encapsulated and firm in consistency. On gross inspection the general substance of the tumor was made up of fibrous tissue, which was gray-white in color and ran in fibrous strands at random through the tumor mass (Fig. 1).

On microscopic examination the tumor was seen to be made up of liver tissue which had been replaced for the most part by collagenized fibrous tissue. Throughout the fibrous tissue mass were found proliferative bile ducts, irregular as to length and configuration, but composed of cells which appeared entirely normal. No evidence of malignancy was seen. It was thought that the tumor represented an adenoma of the liver with marked fibrous displacement, or, as termed in the recent literature, a hamartoma of the liver (Fig. 2).

The child made an uneventful postoperative recovery and is well at the present time.

DISCUSSION

We feel that this case represents a congenital tumor of the liver, fulfilling the criteria necessary to classify it as an adenoma of the bile duct type, or hamartoma.

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Fig. 1.—Cut surface of gross section, hamartoma of liver.

Fig. 2.—Microscopic appearance of hamartoma.

It is difficult to conceive of cirrhosis as the etiologic agent. These tumors could well represent an embryonic accessory liver segment or liver bud which has grown in a disorderly fashion.

CONCLUSION

1. A case of successfully resected adenoma or hamartoma of the liver occurring in a 22-month-old female infant has been presented.

2. It is thought that such tumors represent a benign growth of congenital origin.

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PHEOCHROMOCYTOMA

A Case Report With Details of the Medical and Surgical Management and Experimental Study

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RECENT advances in the diagnosis of pheochromocytoma have led to an increased awareness of the clinical characteristics produced by this relatively rare tumor. The dramatic nature of the symptoms produced and the physiologic complications during treatment have been the stimulus for many reports, particularly recently. The problems of treatment remain, however, and it is felt that the following detailed case report may contribute to a general plan of surgical management.

CASE REPORT

This was the first admission of a 34-year-old white married mechanic with the complaints of acute attacks of abdominal pain, throbbing headaches, and perspiration for 5 years. During this period, typical attacks consisted of severe throbbing frontal headaches, sweating, a feeling that the eyes were going to pop out, blurring of vision, "seeing stars," nausea, vomiting, weakness, palpitation, and retraction of the scrotum. These episodes occurred 1 to 2 times a month with increasing frequency, particularly when the patient was overworked, overfatigued, or after an evening of drinking. The attacks could be elicited regularly by elevating his legs, while working on his back, lying on his left side, and by sexual intercourse. The attacks lasted 5 minutes to 8 hours, usually 1 to 3 hours, and could be aborted occasionally by promptly lying down and resting. The episodes were followed by a sense of extreme fatigue and weakness that often lasted for several hours.

The patient was admitted to the Warsaw Hospital, Warsaw, N. Y., July 12, 1949. During a spontaneous attack his blood pressure rose to 280/120. Intravenous histamine (0.25 mg.) produced all the above signs and symptoms. A cold pressor test caused the blood pressure to rise only 16 mm. of mercury. An insulin tolerance test was normal.

The past history was noncontributory except that the patient had had asthma since childhood and was discharged from the Army because of asthma. Shortly after discharge his asthma disappeared coincident to the onset of the previously described hypertensive attacks. In 1945 the patient had syphilis with a positive Wassermann and was treated with penicillin for 1 month.

The patient was well nourished and in excellent physical condition. Examination of the eye grounds revealed a slight degree of arteriosclerosis with some narrowing of the arteries and early arteriovenous nicking. The heart and lungs revealed no abnormalities and resting blood pressures were consistently in the range of 120/70. The blood Wassermann was negative. The white blood cell count, the red cell count, hemoglobin, blood nonprotein nitrogen, carbon dioxide combining power, serum chlorides, sodium, potassium, total proteins, albumin-globulin ratios, and urinalyses were within normal limits. The basal metabolic rate was −2 per cent. A fasting blood sugar was 142 mg. per cent the morning after an attack, and when repeated during a resting interval was 86 mg. per cent. A metastatic x-ray survey was negative. An intravenous pyelogram revealed distortion of the upper cycles of the right kidney with normal function.

Perirenal air insufflation* revealed a large tumor in the region of the right adrenal gland. Air insufflation on the left revealed no abnormalities. An electrocardiogram and ballistocardiogram were normal during resting intervals.

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*By Dr. John Benjamin.

A hypertensive attack was induced in the Ballistocardiogram Laboratory. Electrocardiogram, ballistocardiogram, respirations, and finger plethysmographic patterns were all simultaneously and continuously recorded. The blood pressure was determined at frequent

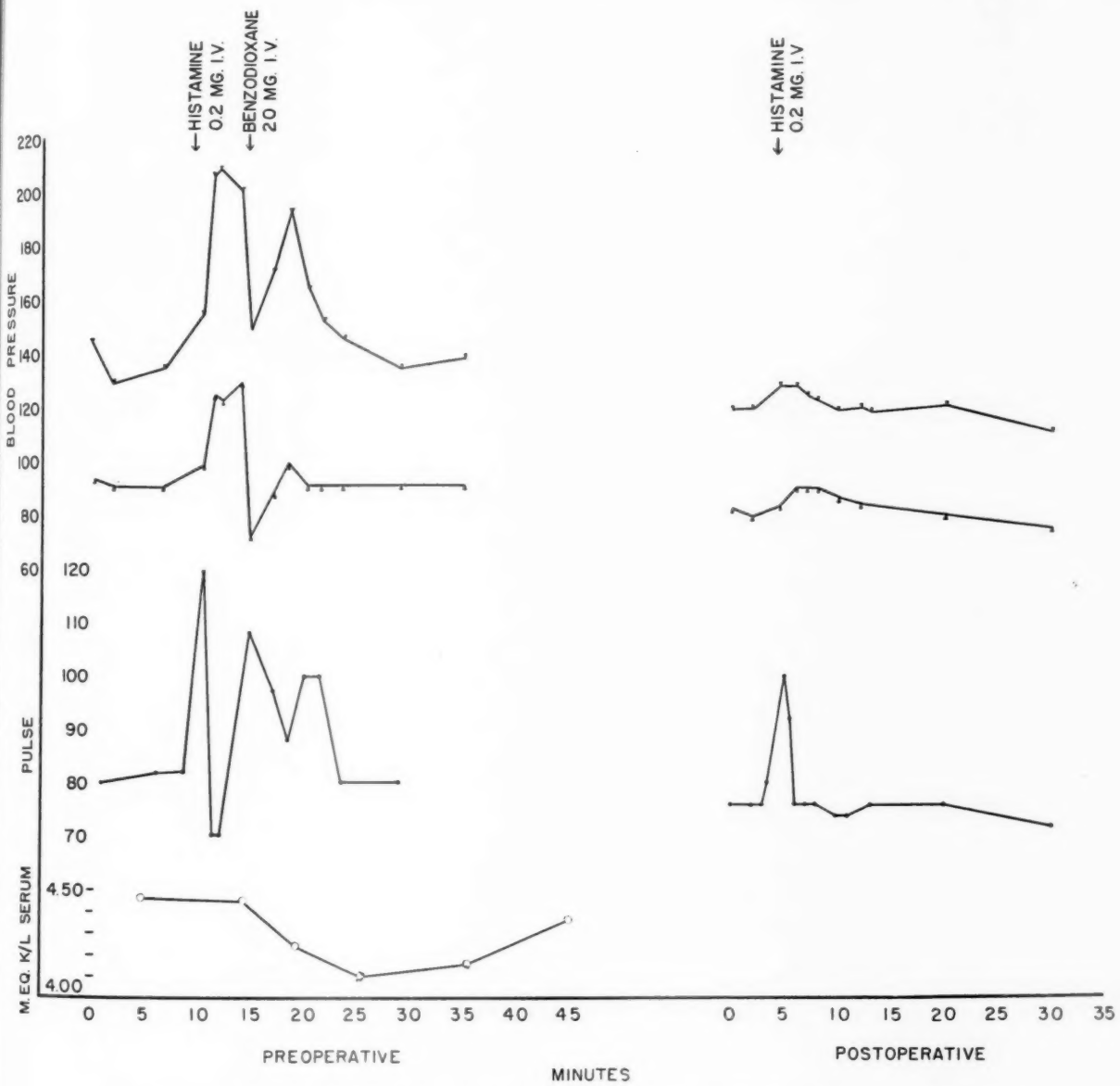

Fig. 1.—Preoperative and postoperative intravenous injection of 0.2 mg. of histamine. Note the effect of Benzodioxane and the resultant brevity of this preoperative hypertensive paroxysm.

intervals. Histamine, 0.2 mg. given subcutaneously, failed to produce an attack. Two-tenths milligram of histamine was then given intravenously (Fig. 1). Less than 1 minute after the injection, the pulse rate increased and the finger volume decreased. The amplitude of the ballistocardiographic complexes increased with deeper K waves than in the control period. The blood pressure rose after the tachycardia was initiated, and when the peak of 210/125 had been reached, the pulse rate was at normal levels. Finger volume remained small.

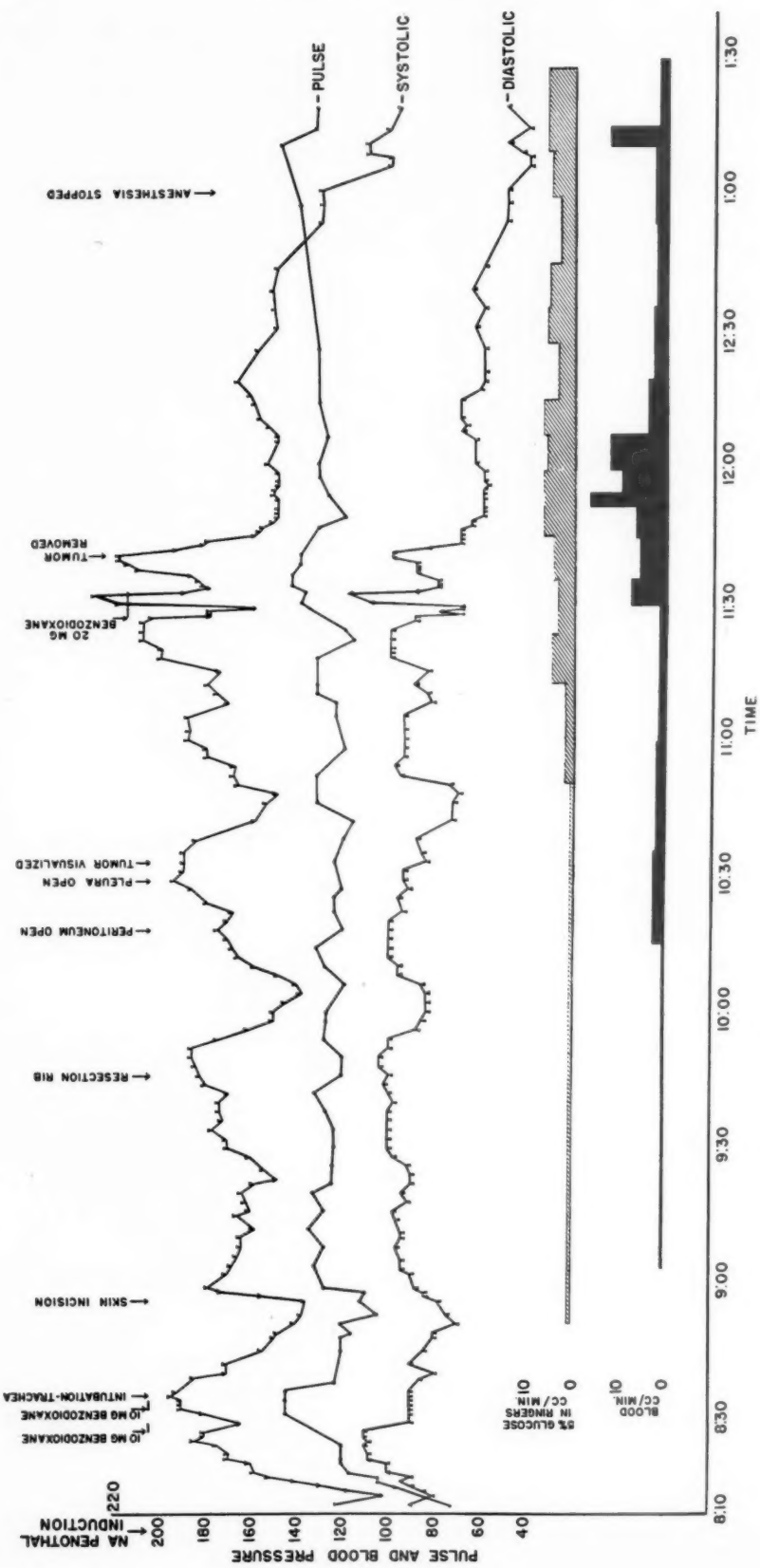

Fig. 2.—Operative record of pulse, blood pressure, intravenous medications, parenteral fluids, and blood.

Four minutes after histamine, Benzodioxane (20 mg.) was given over a period of 1 minute. There was a prompt reduction of blood pressure and increase of finger volume. As the pressure fell there was an increase in the pulse rate and the finger volume again decreased. This was followed by a secondary rise in blood pressure, which promptly fell again. A third less striking increase in pulse rate occurred, but without a rise in pressure. Seven minutes after Benzodioxane, all patterns appeared as they did in the control period. A similar test the following day produced the same pattern.

Repeated 24 hour urine samples were collected with more frequent collections before and after induced paroxysms. Sodium and potassium output was determined for each of these periods (Table I).

TABLE I. URINARY EXCRETION OF SODIUM AND POTASSIUM DURING RESTING PHASES AND HYPERTENSIVE CRISES

PERIOD	TIME (HOURS)	C.C. URINE PER HOUR	MEQ. NA PER HOUR	MEQ. K PER HOUR
1	24	85	6.9	2.5
2	24	49	5.9	2.0
3	24	78	5.3	3.4
4	22.2	96	7.5	2.7
5*	1.83	268	15.1	6.9
6	22.4	120	6.2	3.2
7A*	1	215	13.6	6.3
B	0.78	366	13.1	9.8
C	1.3	181	5.8	4.8
8	24	45	5.6	2.6
9	24	78	6.4	3.9

*0.2 mg. histamine intravenously

The patient was prepared for operation with intravenous lipoadrenal cortical extract, 5 c.c., and 10 mg. of desoxycorticosterone. At operation a right thoraco-abdominal incision was used with resection of the eleventh rib. The liver was mobilized by incising the right triangular ligament and was moved medially. A large tumor measuring 10 cm. in its greatest diameter was exposed above the right kidney. The adrenal cortex was stretched out and adherent to the inferior lateral surface of this tumor, giving it a yellowish color in the area. The tumor was excised after very gentle manipulation and meticulous control of its multitudinous blood vessels from the diaphragm, vena cava, and the right renal pedicle. Benzodioxane and blood were given as indicated in the chart during the operation. The details of the medical management during the operation may be seen in the accompanying chart (Fig. 2). Postoperatively the patient had an uneventful recovery. There was no immediate postoperative period of shock.

The patient was discharged and has been seen in follow-up during the past 10 months with no recurrence of symptoms of a hypertensive attack. A repeated histamine test in the office 2 months ago revealed no significant rise in blood pressure and may be compared with a preoperative histamine test in Fig. 1. The patient has had a recurrence of the asthma. This is hard to evaluate in degree, as he has received increased military disability because of these symptoms.

The tumor mass weighed 360 grams and measured 10 by 9 by 6.5 cm. It was soft and semifluctuant, with a glistening capsule that contained many large blood vessels. The cut surface revealed a cystic center which contained about 75 c.c. of dark blood. The wall varied from 1 to 2.5 cm. in thickness and had a firm yellowish external surface. Microscopically there were diffuse growths of large pale spindle and oval cells from the adrenal medulla containing chromaffin pigment (Fig. 3). These varied markedly in size and shape and occurred in sheets and nests in irregularly sized compartments of fibrous tissue. Throughout all areas there was diffuse hemorrhagic edema and large areas of necrotic tissue. Very little adrenal cortical tissue was seen.

DISCUSSION

The method of using Benzodioxane in this case, we believe, was quite helpful. When given very soon after the development of an attack produced by histamine, it may afford a safer opportunity to observe the physiologic effects of a precipitous rise in blood pressure and when effective may give control of spontaneous hypertensive crises. There is always some hesitancy regarding the diagnostic use of histamine intravenously when a paroxysm is expected. With such a combined use of these 2 drugs some of the inherent dangers may be lessened. Certainly there is abundant evidence that an attack of sudden great hypertension may be fatal.[1, 3, 5, 10-12] During the operative procedure Benzodioxane was effective in controlling the extreme rises in blood pressure.

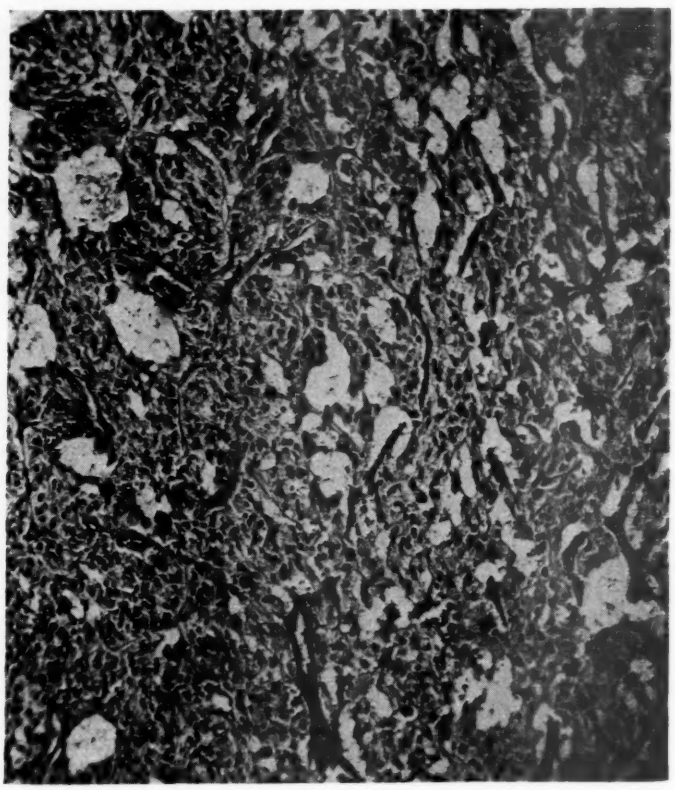

Fig. 3.—Microscopic appearance of the excised pheochromocytoma (×100).

Cahill[2, 3] mentioned an elevated serum potassium in several cases. The reasoning behind this is not recorded, and in the only cases[7, 8] noted in the literature where the potassium level is specifically mentioned it is said to be normal. Wells and Bowman[15] suggested that the serum potassium might rise during an attack. Repeated serum potassium determinations for this patient ranged from 4.5 to 4.7 meq. per liter. (Normal for this laboratory is 4.0 to 5.0 meq. per liter.) Also, serum sodium was within the normal range of 130 to 140 meq. per liter. The level of serum potassium was fol-

lowed during an induced attack terminated by Benzodioxane (Fig. 1). A fall within 5 minutes was observed returning to control levels in 2 hours. These findings are in accord with the effects of injection of epinephrine as reviewed by Fenn.[4] "On injection of adrenalin the potassium first rises and then falls in the blood. The initial rise is so transitory that it can be only detected in man during the first thirty to ninety seconds after injection." The explanation of this rise is not clear but seems connected in some way with the release of liver glycogen and muscle activity.

Of interest was the excretion of sodium and potassium and the urinary output during an induced attack. Twenty-four hour and pre- and post-paroxysmal urine sample analyses are tabulated in Table I. The data, although not conclusive, suggest that the increased output of sodium and potassium reflects increased urinary output. Hyman and Mencher[9] reported decreased urinary output during an attack, which is in keeping with the findings of Smith.[14] The attacks observed in this patient were of short duration, and, therefore, these values probably reflect the period following as well as during the episode. The results suggest, however, that some alteration in kidney function does occur that is related to the hypertensive paroxysm. Our findings are mentioned with the hope that the changes may be clarified.

SUMMARY

A patient with a large pheochromocytoma of the right adrenal is presented, with cure by surgical excision. The details of the medical and surgical management are recorded. Benzodioxane was a useful adjunct in diagnosis, experimental study, and in surgical treatment. Certain studies carried out during spontaneous and induced hypertensive crises are recorded.

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Editorial

The Physician's Professional Status in Army Medicine

Due to the number of questions directed toward the Surgeon General, United States Army, relative to the draft of physicians and the recall of former officers in the Medical Corps, it would seem indicated that a further communication be directed to the physicians of the country. Public Law 779, commonly known as the "Doctor's Draft Law" is now well known to all, but various implications and questions have arisen from it. At the outset let us make it clear that this law was passed by Congress at the request of organized medicine and then passed on to the armed forces for utilization. The Army does not legislate laws nor establish draft policy. The Army, Navy, and Air Forces are to obtain the bulk of medical man power from Priorities 1 and 2 as established, and implemented by executive order from the President and by directives from the Director of Medical Services, Office of the Secretary of Defense. As these categories are exhausted, further groups will be affected. Since many of the ASTP and V-12 program physicians are now in residency training, two things will perforce take place. One is the reduction of hospital resident staffs by approximately 50 per cent of those who are in Priorities 1 and 2, and the other is the influx into the services of partially trained men.

It is obvious, or should be, that all the requirements of the forces cannot be satisfied through the draft alone. The staffing of the Army hospitals will require a certain number of qualified physicians not immediately obtainable through the draft, such as Chiefs of Service, Chiefs of Section, Consultants, and so on. These must be procured through the reserves, or through others who have experience both civilian and military, and must be procured with a definite purpose and post in mind. It is contemplated that such personnel will be called from time to time to active duty for specific positions, and will be requested in a definite manner as to grade, specialty, and qualification, and will be utilized according to their professional training. No great exodus from civilian communities will take place from such procurement. At this time no general blanket recall of reserves is planned or authorized.

For the past three years the Army has maintained a splendid graduate training program, through which it has prepared many medical officers for more skilled work. The results of this program have been well illustrated by the exceptionally fine record being compiled in Korea. When the Far East incident began, a great demand was made upon the regular medical officer. Virtually without exception, the entire Medical Corps of the Far East was made up of regular personnel. Such physicians as were called upon to return were utilized to replace the regulars in this country. But we must realize that with the continuation of the conflict, many civilian practitioners must be called upon to serve overseas.

It is the intent of the Army Surgeon General at present that medical officers will be utilized in the field of their professional training, either directly with patients, or in a supervisory capacity. It should be made clear, however, that in the chaos which is war, some officers must serve in capacities not especially to their liking for temporary or indefinite periods. Furthermore, the incompletely trained and uncertified D category medical officer cannot expect in many instances to be utilized in a specialty, but will be placed in that duty where needed. The day has passed when "surplus" officers will be used to inspect latrines, carry on housekeeping details, and the like. The Medical Service Corps, and other groups, will perform these tasks. It is apparent that some physicians must function in staff level, in command positions, for supervision of services, and must perform other medical administrative tasks. The doctor who claims that even this is unnecessary is indeed unrealistic, for in no other way can medical supervision and work be carried out. Let us examine the data on doctors of medicine in the United States as of Aug. 1, 1947. Of the 197,605 registered physicians at that time, only 74 per cent were in private practice, while the remaining 26 per cent were in some administrative capacity. For example, 4,400 doctors in the United States, or 2 per cent of the total, were employed by insurance companies, industry, and other concerns in administrative positions. Doctors further contributed 2,654, or 1 per cent of the total, as hospital superintendents. How then can it be said that the use of doctors in administrative capacity is peculiar only to the armed forces?

To those who complain that field units are medically overstaffed, let us point out that the ratio of physician to troops has now dropped approximately 50 per cent. The staffing of field medical units is the subject of continuing study and the number of medical officers will be reduced further if this can be done without jeopardizing the welfare of our men. For the most part Medical Service Corps officers and specially trained enlisted men perform the early forward work on casualties, maintain the small dispensaries, and relieve the physician of nonprofessional administrative tasks. As recently stated in regard to 4-F's in general, it must certainly be admitted that the 4-F doctor, in the absence of major organic disease, is capable of carrying out military duties in a military hospital on a limited service basis, especially if he is able to carry on a full-time practice in his home community. Patriotism and sense of duty can be possessed by the physician as well as by his former patient now serving in the combatant forces of his country.

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Recent Advances in Surgery

CONDUCTED BY ALFRED BLALOCK, M.D.

ANOMALIES OF THE PULMONARY VESSELS AND THEIR SURGICAL SIGNIFICANCE

WITH A REVIEW OF LITERATURE

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WITH the technical advancement in resective surgery of the lung, considerable data have been recorded over the past 10 years on the surgical anatomy of the pulmonary circulation. During the first part of this period most of the observations were published by surgeons.[4-9] They were interested chiefly in determining the usual arrangement of the structures at the pulmonary hilus and other details of the anatomy of the lung which were of importance to the surgeon in the performance of individual dissection of the hilar structures.

In 1945, anatomists Boyden[3] and Appleton[1, 2] published their papers on the blood vessels of the lungs independently. These and subsequent papers from the same laboratories are comprised of statistical findings from a significant series of dissections, and they are the most complete reference articles on the subject to date.

These researches have revealed many variations of the pulmonary blood supply, but the authors did not discuss particularly the abnormal connections between the systemic and pulmonary circulations, since these aspects were not pertinent to their reports. The literature on anomalous venous drainage of the lung has been reviewed and analyzed by Brody[74] and later by Brantigan.[73] In the past, reports on anomalous arterial communications and abnormal systemic arteries to the lungs have been for the most part individual and sporadic. Recently, however, they have been more frequent, due to the marked increase in the practice of extirpative surgery of the lungs.

Presented as follows is a statistical review of the literature on arterial communications between the greater and lesser circulations, with comment on the significance of these anomalies. Illustrative cases are also reported.

ANOMALIES OF THE ARTERIAL PULMONARY CIRCULATION

These cases from the literature represent either abnormal communications between the greater and the lesser circulations, or unusual arterial vessels from the systemic circulation to pulmonary parenchyma. Although a careful search has been made to collect examples of each type of anomaly, we realize that some

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suitable reports probably have been overlooked. Many reports are not included here because of insufficient data concerning the vascular arrangements. In fact, it is unfortunate that some of the authors of the case histories listed in the following tables were not more precise and complete in their descriptions.

This series is presented, therefore, in an attempt to provide some order in our concept of these anomalies as well as to provide a basic arrangement for the subsequent addition of other cases that have been published or will be presented in the future.

In order to segregate the cases into important categories, the divisions have been made according to the major vessel of the systemic circulation from which the anomalous artery originated (Fig. 1). The vessels associated with aberrant pulmonary lobes are considered as a separate entity.

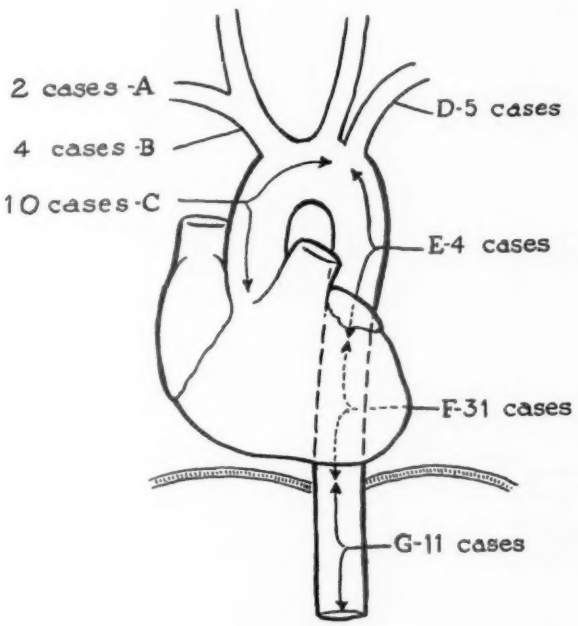

Fig. 1.—This illustration of the heart and great vessels demonstrates the frequency of anomalous pulmonary arteries arising from different segments of the systemic circulation in these collected cases. *A*, from right subclavian; *B*, from innominate artery; *C*, from ascending aorta and aortic arch proximal to left subclavian artery; *D*, from left subclavian artery; *E*, from descending thoracic aorta at or above the level of the pulmonary hila; *F*, from descending thoracic aorta or its branches below the hila; and *G*, from abdominal aorta or its branches.

Aberrant Lung Tissue Above the Diaphragm

These 19 cases are a fraction of those reported. Omissions have been made, however, due to the lack of information about the arterial blood supply to the aberrant pulmonary tissue. Most of these anomalies were seen in newborn infants, but the oldest patient was 49 years old.[26] Rarely did the lesion by itself account for the death of the patient.

Eighty-nine per cent of the aberrant lobes were between the lower lobe of the left lung and the diaphragm. One lobe on the left was found posteriorly at the level of the ninth rib.[28] In another case this anomaly was adjacent to the right diaphragm.[25] Cockayne and Gladstone[12] found 2 accessory lobes in

TABLE I. ABERRANT LUNG TISSUE ABOVE THE DIAPHRAGM

AUTHOR	AGE	SEX	POSITION	ARTERIAL SUPPLY	VENOUS DRAINAGE	CAUSE OF DEATH	ASSOCIATED ANOMALIES	MEASUREMENTS OF ABERRANT TISSUE
Rokitansky[21] (1861)	3 mo.	F	Between left lower lobe & diaphragm	2 arteries from aorta near 10th intercostal arteries	One vein emptied into azygos vein			
Rektorzik[20] (1861)	14 days	F	Between left lower lobe & diaphragm	One artery 2 cm. long and with diameter of renal artery from aorta near 10th intercostal artery	One vein emptied into hemiazygos vein	Peritonitis		4 × 2¼ × 1 cm.
Ruge[22] (1878)	Newborn		Between left lower lobe & diaphragm	One artery from 7th intercostal artery				
Humphry[15] (1884)	1 yr.		Between left lower lobe & diaphragm	One artery from descending aorta	One vein drained to hemiazygos vein	Generalized tuberculosis		2 × 1 inches
Huguenin & Sorel[14] (1888)	16 mo.	F	Between left lower lobe & diaphragm	One artery from a lower intercostal artery or aorta	One vein drained to hemiazygos vein	Croup		53 × 40 × 29 mm.
Springer[26] (1898)	49 yr.	F	Between left lower lobe & diaphragm	One artery 2 mm. in diameter from aorta just above diaphragm	2 veins drained to hemiazygos vein	Heart failure		3 × 2 × 1 cm.
Quensel[19] (1900)	5 yr.	F	Between left lower lobe & diaphragm	2 arteries, one 5 mm. in diameter from aorta between 7th and 8th intercostal arteries	One vein drained to hemiazygos vein	Miliary tuberculosis		
Voisin[27] (1903)	4 yr.	M	Between left lower lobe & diaphragm	2 arteries from aorta at level of 10th & 11th intercostal arteries	One vein drained to hemiazygos vein			5.7 × 4.3 cm.

Lewisohn[16] (1903)	4 mo.		Between left lower lobe & diaphragm; thin pedicle to posterior thoracic wall	One artery, 2.2 cm. long, almost diameter of renal artery, from aorta		Meningitis	4 × 3 × 1.7 cm.
Muus[18] (1904)	1 day	M	Between left lower lobe & diaphragm; short, thick pedicle to diaphragm posteriorly	One artery from region of adrenal gland		Cyanosis	Size of a walnut
Beneke[10] (1905)	Newborn		In left chest; adherent to stomach	One artery from abdominal aorta via aortic hiatus; sent one branch to diaphragm	One vein drained to portal vein along greater curvature of stomach	Enormous diaphragmatic hernia	The aberrant lobe was as large as the other lobes on the left
Simpson[25] (1908)	Full term fetus		Between right lower lobe & diaphragm; long narrow pedicle to mediastinum	One retroesophageal artery, size of carotid artery, from aorta at level of D 10	Several veins draining to esophageal and azygos veins	Right lung normal but small	2 × 1¼ inches
Sachs[23] (1912)	Newborn	F	Between left lower lobe & diaphragm; 1.5 cm.; pedicle to diaphragm	One artery, probably from phrenic artery	One vein to hemiazygos vein	Anasarca	Pyramidal shaped; 6 × 4 × 4 cm.
Von Mayenburg[28] (1914)	5 yr.	M	In left posterior chest at level of 9th rib	One artery from descending aorta	One vein to left pulmonary vein	Appendicitis & peritonitis	40 × 28 × 3 mm.; rhomboid shaped
Cockayne & Gladstone[12] (1917)	Infant	F	2 aberrant lobes between left lower lobe & stomach; each lobe had a pedicle to aorta 1.75 long	2 arteries from aorta to upper aberrant lobe; one artery to lower aberrant lobe	One vein drained to hemiazygos vein	Died shortly after birth	Diaphragmatic hernia	Upper lobe pear-shaped, 3.4 cm. in diameter; lower lobe fan-shaped, 4 × 4.5 × 1.2 cm.

TABLE I—CONT'D

AUTHOR	AGE	SEX	POSITION	ARTERIAL SUPPLY	VENOUS DRAINAGE	CAUSE OF DEATH	ASSOCIATED ANOMALIES	MEASUREMENTS OF ABERRANT TISSUE
Marfori[17] (1935)	11 mo.	F	Between left lower lobe & diaphragm	Several arteries from aorta just above diaphragm	One vein drained to hemiazygos vein			Globular
Carter & Osborn[11] (1936)	6 days	F	Between left lower lobe & diaphragm; pedicle to diaphragm	One artery from aorta at level of celiac axis, through diaphragm to pedicle		Cyanotic death	Wide patent ductus; patent foramen ovale	
Sheidegger[24] (1936)	Newborn	M	Between left lower lobe & diaphragm; thick fibrous pedicle to diaphragm	One artery from descending aorta; another smaller artery from left kidney	One vein drained to hemiazygos vein		Left lung failed to expand	52 × 45 × 38 mm.
Davies & Gunz[13] (1944)	Newborn	M	Between left lower lobe & diaphragm; attached to diaphragm	One artery from branch of aorta near diaphragm	One vein to azygos vein		Large left diaphragmatic hernia	1½ × ¾ inches

1 individual. The aberrant tissue was always completely invested with pleura, which was usually free of adhesions. Sometimes the lobes had vascular pedicles which were covered with pleura. On other occasions they were adherent to the diaphragm at the source of vascular supply.

The arterial blood supply to the aberrant lobe came from the descending thoracic aorta in 14 cases. It originated from the abdominal aorta on 2 occasions and once from an intercostal artery, the phrenic artery, and from the region of the adrenal gland respectively. There were 4 instances of multiple arteries to aberrant lobes. Those arteries coming from the thoracic aorta usually arose at the level of the tenth intercostal space. In Simpson's case,[25] where the aberrant lobe was on the right, the nutrient artery arose from the lower thoracic aorta, and coursed behind the esophagus to reach its destination.

The venous drainage of 48 per cent (9 cases) of these lesions emptied into the hemiazygos vein. In 4 cases the drainage was into the azygos vein; 3 of these were cases of left-sided lobes. Von Meyerding[28] noted a single vein coursing to the left pulmonary artery. In Beneke's case,[10] which was associated with a diaphragmatic hernia, a vein drained ultimately into the portal system.

The bronchi of these aberrant pulmonary lobes did not communicate with the rest of the bronchial tree in any instance. Associated diaphragmatic hernias were noted in 3 cases. In this series the heart was almost always normal.

Airless lung tissue was noted on all pathologic sections. Bronchi were observed which were lined with columnar and sometimes ciliated epithelium. On occasions the specimens contained cysts resulting from distention of the bronchioles. Simpson's[25] case demonstrated many of these features with widespread degeneration. The anomaly reported by Humphry[15] showed tuberculous caseation, illustrating that aberrant lung tissue is also susceptible to systemic disease.

Cockayne and Gladstone[12] and Davies and Gunz[13] have written excellent discussions on the embryology of this anomaly.

Aberrant Lung Tissue Below the Diaphragm

These cases are similar to those outlined in Table I in many respects. The average age of these patients, however, is older, and the aberrant tissue is usually adherent to the adjacent structures. Valle[29] was the first to discover this anomaly at operation.

Anomalous Pulmonary Blood Supply From the Descending Thoracic Aorta

The average age in this series was about 17 years; the oldest was 65 years of age. In 10 of the 17 cases reported, the anomalous vessels went to the left lower lobe. The lower lobe of the right lung was supplied on 3 occasions and the left upper lobe once. Two cases had aberrant vessels going to both lower lobes.

From the anatomic description of 7 of these cases it can be concluded that the origins of the anomalous arteries were at the level of the tenth thoracic vertebra. In 3 instances these vessels arose at the level of the hila of the lungs.

TABLE II. ABERRANT LUNG TISSUE BELOW THE DIAPHRAGM

AUTHOR	AGE	SEX	POSITION	ARTERIAL SUPPLY	VENOUS DRAINAGE	CAUSE OF DEATH	ASSOCIATED FINDINGS	MEASUREMENTS OF ABERRANT TISSUE
Vogel[30] (1899)	47 yr.	M	Just to right of left kidney; upper portion in aortic hiatus of diaphragm	Two arteries from thoracic aorta at the level of 10th interspace	One vein draining to hemiazygos vein	Carcinoma of stomach	Numerous adhesions	Saucer-shaped
Vogel[30] (1899)	28 yr.	F	Between left diaphragm and kidney	Artery from celiac axis; 4 branches from left inferior phrenic artery	Veins draining to suprarenal vein		Numerous adhesions	Pyramidal; 4.5 × 4 cm.
Valle & White[29] (1947)	9 mo.	M	Beneath left diaphragm	Several large arteries from aorta		Vomiting before death; cause of death unknown	Thinned diaphragm	4 × 3 × 2 cm.

The anatomic accounts of the remainder of the cases are not specific enough to denote the precise level of origin. In most instances these vessels traveled from the aorta to their pulmonary destination via the pulmonary ligaments. In those cases where the vessels arose at or above the bifurcation of the trachea, hypertrophied bronchial arteries must be suspected.

There were 6 cases in which there were no pulmonary arteries from the lesser circulation to the abnormally supplied segments of lung. In the majority of the remaining cases the presence or absence of a dual pulmonary arterial circulation was not stated, although in a few there were definitely concomitant branches from the pulmonary artery.

Pulmonary atresia or hypoplasia with associated severe cardiac anomalies was noted on 3 occasions. It is assumed from the descriptions of 9 cases that the hearts were normal. In the remaining cases the data presented are insufficient to allow adequate appraisals of the hearts.

In 7 cases inflammatory processes were demonstrated in the lobes that were supplied by systemic arteries. The underlying pathology was bronchiectasis, congenital pulmonary cysts, or abscesses of unknown etiology. Congenital malformation of the bronchopulmonary segments were suspected in almost every instance. All of the 7 cases were noted at operation and there were 2 deaths directly attributable to hemorrhage following transection of the anomalous arteries.[35, 36]

Case 1. Malformation of the Heart, Constrictive Vascular Ring Due to Bilateral, "Patent Ducti Arteriosi," and Three Left Pulmonary Lobes.—

Illness at time of admission: A. M. C. (P. H. No. 922381), a 3-month-old female infant, was admitted to the Babies Hospital, New York, N. Y., on Sept. 10, 1948, with the chief symptom of intermittent labored breathing following her discharge from the hospital at the age of 2 weeks. Roentgenograms taken at that time showed congestion of the right lung. 2 weeks prior to admission to the hospital, cyanosis was noted for the first time when the child cried.

Physical examination: Temperature was 98.6° F., pulse 140, and respirations 66. The infant was obviously malnourished and appeared chronically ill. The skin was pale without cyanosis. There was slight subcostal retraction on inspiration. Both sides of the upper chest were dull to percussion and increased breath sounds were heard over these areas.

The heart sounds were of good quality and the cardiac rate was rapid. There was a blowing systolic murmur in the second and third interspaces to the left of the sternum which was transmitted to the back.

Laboratory data: The blood hemoglobin was 10.5 Gm. The red blood cell count was 3,500,000 and the white blood cell count was 7,700 with 34 per cent polymorphonuclears.

Roentgenograms of the chest showed shadows of increased density in both upper lung fields, most markedly on the right (Fig. 2). The right cardiac border was not sharply defined, but the left ventricle was enlarged. Roentgenographic examination following a barium swallow showed an oblique indentation of the esophagus posteriorly at the level of the fifth rib anteriorly. This defect pointed toward the left shoulder. A tracheogram with the aid of Lipiodol showed no evidence of pressure on the trachea, but the right main bronchus was obviously compressed from above (Fig. 3). The infant was dyspneic during these examinations, especially when she lay on her abdomen. The clinical impression at this time was that the patient had cardiac and aortic arch anomalies with a constrictive vascular ring.

Course: On Oct. 8, 1948, a left thoracotomy was performed through an anterior incision. It was evident that the child had a right aortic arch. A retroesophageal artery was seen coursing to the left pulmonary artery. It was thought that this vessel arose from the right

TABLE III. ANOMALOUS PULMONARY BLOOD SUPPLY FROM THE DESCENDING THORACIC AORTA

AUTHOR	AGE	SEX	ANOMALOUS ARTERIAL SUPPLY	CAUSE OF DEATH	ASSOCIATED ANOMALIES	OTHER FINDINGS
Huber[37] (1777)	2 yr.	F	Large artery from thoracic aorta at level of D 10 entering substance of right lower lobe; it sent branches to esophagus and bronchi			
Meckel[39] (1820)	9 mo.		Artery 9 mm. in diameter from aorta just above diaphragm to basal border of left lower lobe			
Hyrtel[38] (1839)	Newborn		Artery from thoracic aorta to left lower lobe		The left pulmonary artery supplied only the left upper lobe	
Weiss, S.[43] (1875)	6 yr.	F	Artery 2 cm. in diameter from aorta at lower border of hilum of lung to left lower lobe	Cyanotic death	Tetralogy of Fallot	
Babington[31] (1866)	9-10 yr.		Artery from aorta bifurcated and sent a branch to right lower lobe and left lower lobe		Two vessels arose from patent ductus arteriosus to right lung; an obstructed branch also arose from ductus to left lung	
McCotter[40] (1910)	65 yr.	M	Artery 7 mm. in diameter from aorta at level of D 10 to right lower lobe via pulmonary ligament	Heart disease (anatomic dissection)	No accompanying vein	Normal parenchyma of right lower lobe
Dickson & Fraser[34] (1914)	4 mo.	M	Artery from descending aorta coursed behind esophagus to left lung	Not clear; vomiting before death	Pulmonary atresia, interventricular septal defect, right aortic arch; large artery from aorta ½ inch above aortic ring to right lung	
Paul, F.[42] (1928)	3 yr.	F	Artery one-half the size of aorta arose from it just above diaphragm to basal portion of left lower lobe via pulmonary ligament	Miliary tuberculosis	Concomitant normal branch of pulmonary artery to left lower lobe	Normal bronchial arteries; Lipiodol injected into anomalous artery showed anastomoses with lesser circulation
Wheeler & Abbott[44] (1928)	29 yr.	M	Artery from aorta behind right main bronchus sent branches to both lungs; smaller artery from aorta distally coursed to left lower lobe	Very cyanotic	Pulmonary atresia, interventricular septal defect, right aortic arch, small patent ductus arteriosus to right pulmonary artery	

Brenner[32] (1936)	8 mo.	F	Artery one-half the size of aorta from right side of aorta at level of 3rd intercostal artery coursed behind esophagus, over right main bronchus to right lung; it branched in the manner of a bronchial artery	Cardiac failure; bronchitis	Ductus arteriosus functionally closed; patent foramen ovale	Heart greatly enlarged; right side greater than the left
Harris & Lewis[36] (1940)	5 yr.	F	Artery as large as celiac axis from aorta 2 cm. above diaphragm to left lower lobe via pulmonary ligament	Operative death		Abscess and bronchiectasis of left lower lobe
Moore[41] (1940)			Artery from aorta 1 inch above diaphragm through dense adhesions to affected lobe	Operative case		Infected cyst of lower lobe
Brown & Robbins[33] (1944)	21 yr.	F	Artery from thoracic aorta to well-fissured posterolateral segment of basal division of left lower lobe	Operative case; left lower lobe lobectomy	The independent segment contained an infected bronchiogenic cyst	Normal branch of pulmonary artery to left lower lobe
Douglass[35] (1948)	15 yr.	F	Artery 6 mm. in diameter from either aorta or intercostal artery to base of left lower lobe	Operative case; left lower lobe lobectomy		Abscess of left lower lobe
Douglass[35] (1948)	22 yr.	F	Artery 4 mm. in diameter from aorta to left lower lobe	Operative case; left lower lobe lobectomy		Abscess of left lower lobe; lobe was densely adherent to paraspinal gutter
Douglass[35] (1948)	20 yr.	F	Artery 4 mm. in diameter from aorta to diseased portion of left lower lobe via pulmonary ligament	Operative case	Injection of anomalous artery showed branches coursing to the cystic area	Infected congenital cyst of left lower lobe
Douglass[35] (1948)	13 yr.	M	Artery 5 mm. in diameter from aorta; it coursed to the left lower lobe between the diaphragm and the parietal pleura	Operative death due to transection of anomalous artery	Multiple congenital cysts of left lower lobe	Dense adhesions between left lower lobe and diaphragm; normal branch of pulmonary artery to left lower lobe

Eight cases of an abnormal artery arising from the aorta close to the diaphragm have been reported by Pryce and associates. All were operative cases for suppurative conditions or pulmonary cysts. (Pryce, D. M., Sellors, T. H., and Blair, L. G.: Intralobar Sequestration of the Lung Associated With an Abnormal Pulmonary Artery, Brit. J. Surg. 35: 18, 1947.)

arch and the diagnosis of an unusual patent ductus arteriosus was considered. No untoward signs or symptoms developed after it was temporarily occluded. Since it was believed that this vessel was the primary cause of the respiratory symptoms, it was doubly ligated and divided. The left innominate and right subclavian arteries were anchored to the sternum by stitches. The remainder of the operation and closure of the wound proceeded uneventfully.

On the evening of the operation the patient became markedly cyanotic and showed suprasternal and infracostal retraction on inspiration. Breath sounds were diminished throughout both lung fields. A tracheotomy was performed with considerable improvement of the child's clinical picture. The following day, however, she again developed cyanosis

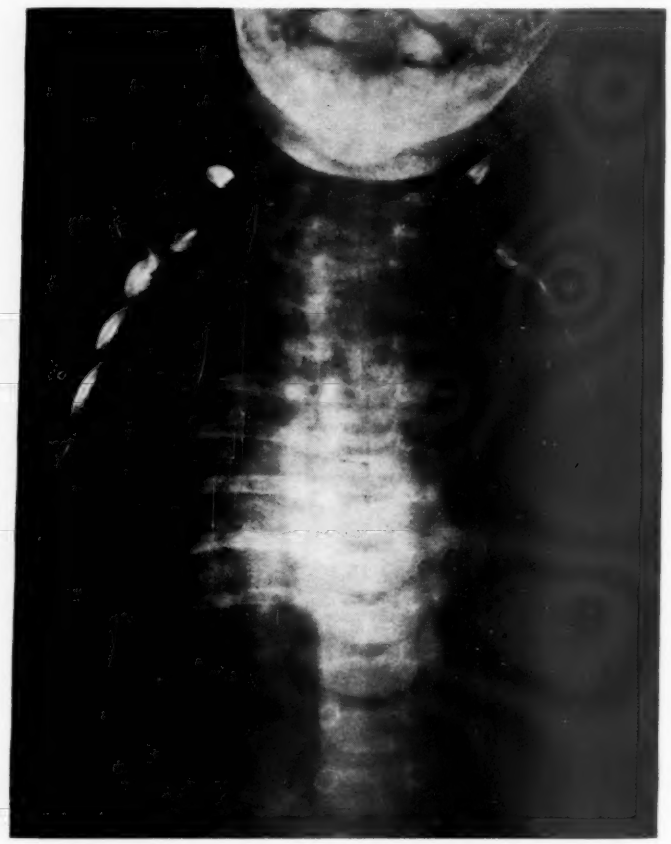

Fig. 2 (Case 1).—Roentgenogram of the chest showing configuration of heart and scattered areas of increased density in both upper lung fields.

and tachypnea which was partially relieved by the administration of oxygen. During the next 24 hours her general condition became worse and death ensued on the fourth postoperative day. The clinical diagnosis was congenital heart disease, patent ductus arteriosus, subglottal edema, and cardiac failure.

Autopsy findings: The post-mortem findings significant to this subject are as follows: The left pleural cavity contained 15 c.c. of bloody fluid, but there was no pneumothorax. The left innominate and the right subclavian arteries were sutured to the sternum.

There was atresia of the pulmonary orifice. The pulmonary artery measured 9 mm. in circumference just proximal to its bifurcation. The branches were hypoplastic. The heart was globular and more prominent on the right. The great systemic veins were not unusual. On examination the interior of the heart, the right auricle and ventricle appeared

dilated and hypertrophied. There was a patent foramen ovale, an interventricular septal defect, and atresia of the pulmonary valve. The pulmonary venous drainage emptied into the left auricle. The left ventricle was hypertrophied and the coronary arteries were normal.

There was dextraposition of the aorta, which was dilated. It measured 4.4 cm. in circumference just above the aortic valve (Figs. 4 and 5). It coursed over and indented the superior aspect of the right main bronchus and descended in the right chest. The left innominate and right common carotid arteries had a common origin 3 cm. above the aortic ring. The former vessel passed anteriorly in front of the trachea. The latter ascended into the neck; 1.2 cm. distal to the orifice of these vessels and on the descending limb of the arch, the right subclavian artery arose and coursed to the right arm. Three large vessels, 2.9 cm. distal to the origin of this vessel, arose from a common orifice. One of

Fig. 3 (Case 1).—Arrows point to pressure defect of right main bronchus as demonstrated by bronchography. Lipiodol has spilled over into esophagus.

these, measuring 2.2 cm. in length and 0.5 cm. in diameter, passed behind the right main bronchus to anastomose with the right pulmonary artery. Another artery passed to the left, behind the esophagus, to connect similarly with the left pulmonary artery. This was the structure that was ligated and divided at operation. The third and smallest vessel passed in front of the esophagus to the left lung. It coursed along the postero-inferior aspect of the left pulmonary artery with which it did not communicate grossly. Three millimeters distal to the orifices of the previously mentioned vessels, a large artery arose from the posterior surface of the aorta, which coursed to the right pulmonary hilum. This was considered to be a hypertrophied bronchial artery. It was 2.4 cm. long and 9 mm. in circumference.

There were 3 left pulmonary lobes and there was widespread pulmonary consolidation bilaterally due to chronic passive congestion.

Comment.—If this infant could have tolerated a retrograde aortogram and cardiac catheterization, valuable information of the pathology and physiology might have been obtained. The surgical division of a probably beneficial abnormal vessel would not have been done. (See discussion.)

Anomalous Pulmonary Blood Supply From the Abdominal Aorta or Its Branches

In this group of 8 cases, the ages of the patients ranged from 2½ months to 40 years.

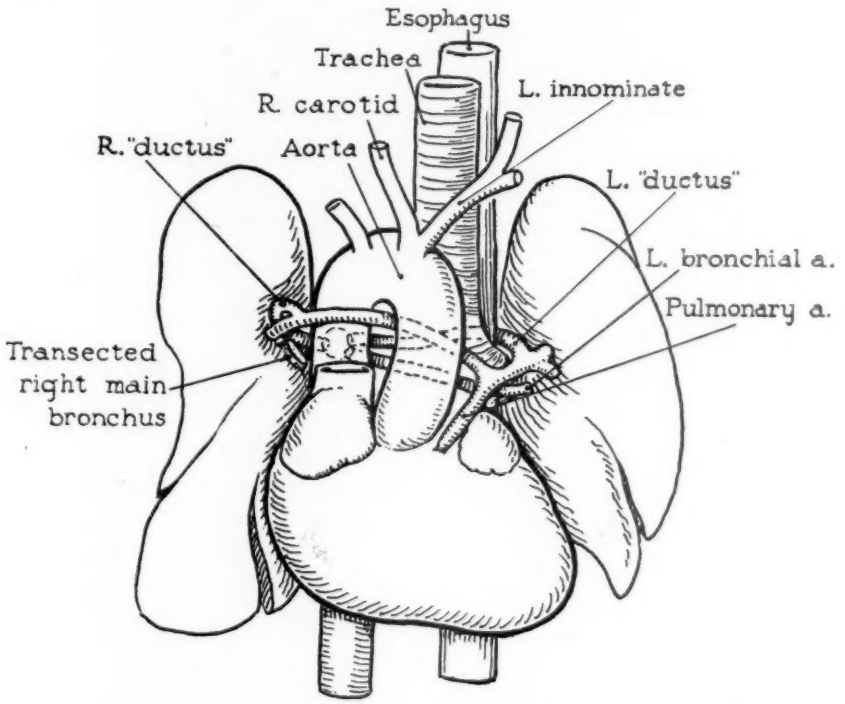

Fig. 4 (Case 1).—Anterior view illustrating heart and great vessels. (See text.)

The anomalous pulmonary arteries from the greater circulation arose either from the abdominal aorta just below the diaphragm, from the celiac axis or from one of the branches of the axis. These vessels transversed the diaphragm through normal or abnormal diaphragmatic hiati. If such an artery entered the thoracic cavity close to the midline, its usual course was then between the leaves of the pulmonary ligament. In 4 instances the lower lobe of the right lung received an anomalous artery, the lower lobe on the left was supplied once and abnormal vessels to both lower lobes were observed on one occasion.

In none of the cases listed in either Tables III or IV was there associated pulmonary venous drainage to the right heart except in the one recorded by Park.[51] In his case a pulmonary vein coursed to the inferior vena cava. There was evidence of congenital bronchopulmonary anomalies in one-half of the

cases listed in Table IV. Only Cioni's[47] case had a severe associated cardiac anomaly. Four cases showed impaired arterial supply from the lesser circulation to the abnormally supplied pulmonary lobes.

Case 2. Anomalous Right Lung Without a Lesser Circulation; Blood Supply Via Small Arteries From Phrenic or Subphrenic Branches; Venous Drainage to Superior Vena Cava.—J. C. (P. H. No. 738199), a 14-year-old Negro boy, was first admitted to the Presbyterian Hospital in 1944 because of fever and a history of hemoptysis. Previously he had been hospitalized elsewhere 3 times for bronchopneumonia since he was 6 months of age. When he was an infant his mother was told that he had a completely collapsed right lung with a cardiac shift to the right. She was reassured that he did not have primary heart disease.

Between hospital admissions, he had frequent upper respiratory infections and a persistent cough which gradually became productive of one-half cup of yellowish sputum daily. Although his activities were not seriously curtailed, he was moderately dyspneic on exertion. One month prior to admission he began to complain of weakness and fatigue. He coughed up a teaspoonful of blood at this time, and again 2 weeks later. For 3 days before entry he had fever and right anterior chest pain.

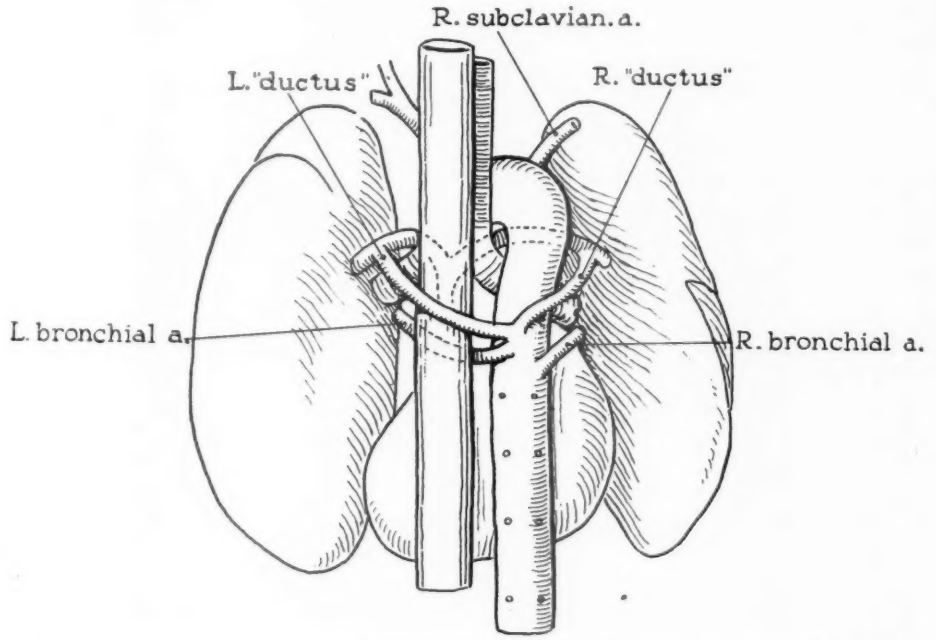

Fig. 5 (Case 1).—Posterior view illustrating heart and great vessels. (See text.)

On his first admission a diagnosis was made of bronchopneumonia on the right and atelectasis of the lower lobe of the right lung with a mediastinal and cardiac shift to that side. A good clinical response to oral sulfadiazine was obtained. Bronchography showed a well-outlined bronchial tree of the right upper lobe, which was overexpanded (Figs. 6 and 7). The right main stem bronchus ended in a blind sac measuring 2 cm. in diameter.

He was readmited in March, 1948, for operative treatment. At this time he was small in stature, but well nourished and well developed. He did not appear ill and there was no clubbing of the digits. There was decreased expansion of the right chest with diminished breath sounds, dullness, and numerous râles on that side. A rough systolic cardiac murmur which was heard in the right second and third interspaces anteriorly disappeared on exercising.

TABLE IV. ANOMALOUS PULMONARY BLOOD SUPPLY FROM THE ABDOMINAL AORTA OR ITS BRANCHES

AUTHOR	AGE	SEX	ANOMALOUS ARTERIAL SUPPLY	CAUSE OF DEATH	ASSOCIATED ANOMALIES	OTHER FINDINGS
Maugars[50] (1802)	7 yr.	M	Artery 5 mm. in diameter from aorta just above celiac axis; it coursed through esophageal hiatus, behind esophagus, and bifurcated, sending a branch each to right lower lobe and left lower lobe	Unknown but not due to anomalies	No branches of normal pulmonary arteries to either lower lobe	Post-mortem injection studies of specimen through the pulmonary artery showed communications with branches of anomalous artery
Eppinger & Schauenstein[48] (1902)	4 mo.	M	One artery from celiac axis to root of rudimentary lobe of right lung; another artery from celiac axis to right upper lobe via pulmonary ligament	Left lobar pneumonia	Upper lobe on right fairly normal; only other lobe was posteromedial, had no proximal bronchial connection, and was hypoplastic; right pulmonary artery was 2 mm. in diameter and sent branches to both lobes	The left pulmonary artery and left lung were larger than normal
Park[51] (1912)	2½ mo.	M	Four arteries from right phrenic artery to inferior portion of right lung		Right pulmonary artery smaller than left; no right pulmonary veins draining to left auricle; one large vein drained blood from right lung into inferior vena cava just above diaphragm	Right lung collapsed and firm; tiny bronchial artery on right; normal pulmonary venous drainage on left
Cioni[47] (1939)	12 yr.	F	One artery 9 mm. in diameter from celiac axis traversed the right diaphragm to the right lower lobe	Pneumonia and cyanosis	Tetralogy of Fallot with valvulitis of the mitral, aortic, and pulmonic valves; interatrial septal defect	
Batts[46] (1939)	40 yr.		Artery 8 mm. in diameter and 9 mm. long arose from celiac axis, passed through right diaphragm near esophageal hiatus to medioinferior portion of right lung	Heat stroke, general paralysis, and empyema		Normal right bronchial arteries; injection studies showed only medioinferior portion of right lower lobe supplied by anomalous artery
Arce[45] (1943)	15 yr.	M	Artery 5 mm. in diameter from systemic circulation below diaphragm to left lower lobe via pulmonary ligament and diaphragmatic adhesion	Operative finding; left lower lobe lobectomy		Bronchiectasis of lower lobe of left lung
Haight[49] (1944)	Two identical cases		An artery in each case the size of a lead pencil traversed diaphragm to supply affected lower lobes		Sides of lesions not mentioned	Congenital cysts of lower lobes

The blood count was normal. Roentgenograms of the chest revealed essentially the same findings as noted previously, except that there was apparent increased aeration of right upper lung field. It was observed for the first time that the xiphoid and lower portion of the body of the sternum were absent. Bronchograms taken outside the hospital shortly before admission showed no change from those taken in 1944. The electrocardiogram was not remarkable and sputum examinations were negative for tubercle bacilli.

A right upper lobe lobectomy was performed on April 1, 1944. The postoperative recovery was satisfactory, and 12 days later he had partial scapulectomy and a thoracoplasty of the Sauerbruch type with removal of the posterior segments of the second to the seventh ribs inclusive. He made a good recovery and was discharged from the hospital on April 22, 1948.

Fig. 6 (Case 2).—Roentgenogram showing a shift of the heart and mediastinum to the right, elevation of the right diaphragm, and intercostal narrowing on the right due to diminished volume of the right lung of long duration.

Pathologic findings: At operation the right pleural cavity was obliterated by exceedingly dense and vascular adhesions. There were several firm bands from the diaphragmatic pleura to the lower surface of the lung containing large vessels measuring 2 to 3 mm. in diameter. With hilar dissection, a nodular stump of the main stem bronchus was uncovered. There were no normal pulmonary arteries or veins on this side. A large vein, however, which was definitely not the azygos, left the single lobe, ran parallel to the bronchus, and emptied into the superior vena cava.

The segment of the right main bronchus which was removed with the specimen measured 2.5 cm. in length. The upper lobe bronchial orifice was at its usual site. Just distal to this, a blind pouch represented the remnant of the main stem bronchus to the

lower and middle lobes. The upper lobe bronchus bifurcated into a relatively normal superior branch and an inferior branch which promptly ended in a blind thin-walled cavity. This measured 1.5 cm. in diameter and did not contain cartilage. Another similar, but smaller pocket was observed leaving the upper branch. The parenchyma of the upper lobe was somewhat emphysematous.

Microscopic examination revealed diffuse emphysema with alveolar rupture throughout the lung parenchyma. The walls of the upper lobe cavities consisted of smooth muscle bundles without cartilage and they had an indefinite epithelial lining. The pouch representing the undeveloped main stem bronchus demonstrated all normal bronchial elements.

Three weeks following discharge from the hospital, the patient had gained 6 pounds and was without complaint.

Fig. 7 (Case 2).—Bronchogram demonstrating termination of the main stem bronchus to the right lower and middle lobes in a blind pouch.

Comment.—This is an example of an anomalous pulmonary circulation that can be puzzling to the surgeon. Obvious bronchopulmonary abnormalities as seen in this case should warrant a search for unusual vessels to the lung.

Case 3. Aberrant Blood Supply to Segment Containing Congenital Pulmonary Cyst.—D. D., a 24-year-old white man, applied for treatment of an intermittently productive cough. He had had no pulmonary signs or symptoms until 4 years previously, when he had pneumonia for 2 weeks. This illness occurred 1 month following an inguinal

herniorrhaphy. Since that time he occasionally had had a cough productive of yellow sputum. During the few months prior to hospital admission the breath and sputum became foul, and during the previous 3 weeks the exudate was streaked with blood. The cough was accentuated when he lay on his left side. As long as he could remember, he had been slightly short of breath on exertion. In spite of this he had served in the United States Marine Corps from 1944 to 1946, during which time chest films allegedly had been interpreted as negative.

The patient appeared healthy. The examination was essentially negative except for the following pathologic findings. Breath was definitely foul. There was dullness at the left base posteriorly, where numerous moist râles were heard. Blood pressure was 130/80.

Fig. 8 (Case 3).—Roentgenogram showing Lipiodol injection of anomalous systemic artery to left lower lobe following resection. (See text.)

Roentgenograms taken shortly before examination showed a cavity in the base of the lower lobe of the left lung behind the heart. Within its thin walls an air-fluid level could be seen.

Because of the evidence of a large amount of retained secretions in the cavity, it was decided to perform a pneumonotomy and then do a lobectomy at a later date. Accordingly, a first-stage drainage was performed. When the second-stage procedure was done 5 days later, the free pleural space was entered with unusual ease. In order to avoid contaminating the pleura, the operation was terminated. During surgery a small incision in the lung caused much more bleeding than expected.

TABLE V. ANOMALOUS PULMONARY BLOOD SUPPLY FROM LEFT SUBCLAVIAN ARTERY

AUTHOR	AGE	SEX	ANOMALOUS ARTERIAL SUPPLY	CAUSE OF DEATH	ASSOCIATED ANOMALIES	OTHER FINDINGS
Jackson[55] (1846)	5 yr.	F	A branch from left subclavian to a large left pulmonary artery; probably a patent ductus arteriosus	Gastroenteritis; history of mild heart disease		Pulmonary artery larger than aorta
Bernard[52] (1846)	Newborn		Large branch from left subclavian to the truncus pulmonalis	Not determined at autopsy	Right-sided aortic arch and descending aorta	
Chevers[53] (1866)	9-10 yr.		Pulmonary artery received blood from branch of the left subclavian artery		Pulmonary atresia; patent interventricular septum	
Meyer[56] (1857)	11 yr.	F	Large artery from left subclavian artery to hilum of left lung		Pulmonic stenosis; ligamentum arteriosus to left subclavian artery 17 mm. proximal to abnormal vessel	
Hewitt[54] (1930)	64 yr.	M	Artery 1.5 mm. in diameter to hilum of left lung behind left main bronchus		No other bronchial arteries to left lung noted	

Following an uneventful postoperative recovery, a lower lobe lobectomy of the left lung was performed for the infected cyst. There was a moderate amount of serosanguinous pleural fluid. The lower lobe was adherent over a localized area adjacent to the pulmonary cavity. A large systemic artery of subphrenic origin measuring 1 cm. in diameter traversed the diaphragm close to the sympathetic trunk. It coursed cephalad in the retropleural space to enter the pulmonary ligament. There was also a pulmonary artery to the lower lobe from the lesser circulation. Two inferior pulmonary veins coursed along the anteroinferior aspect of the main stem bronchus. There were several small veins adjacent to the anomalous artery.

The normal pulmonary arteries were ligated and divided first, then the pulmonary veins, and finally the anomalous artery was seen. Following ligation of this last vessel, the systemic blood pressure exceeded 200 mm. of mercury without any other obvious physiologic changes. The pressure gradually returned to normal. The postoperative period was uneventful.

Special studies: Injection of the anomalous vessel with Lipiodol showed it to supply the posteromedial basal segment of the lobe which was partially occupied by the congenital cyst (see Fig. 8). The divisions of this artery did not freely communicate with the pulmonary arterial branches from the lesser circulation in the adjacent parenchyma of the lung. There was no apparent artery from the pulmonary circuit entering the diseased segment.

Comment.—A solitary cyst of the lung that is close to the diaphragm, especially on the left, is probably congenital. With such an abnormality, anomalous pulmonary blood supply should be suspected. The increased capillary blood pressure in the diseased pulmonary segment caused unusual hemorrhage when it was incised.

Anomalous Pulmonary Blood Supply From the Left Subclavian Artery

All of the individuals represented in these cases died before adolescence except 1, who reached the age of 64 years.[54] Each anatomic diagnosis was different so that it is difficult to make any generalities. In some of these cases an unusual patent ductus arteriosus might be postulated. Meyer's case[56] is of interest, however, in that he described such an anastomotic vessel concomitant with a ligamentum arteriosum which also went to the left subclavian artery.

Two of these cases had associated hypoplasia of the pulmonary arteries. A right-sided aorta may be present.

TABLE VI. ANOMALOUS PULMONARY BLOOD SUPPLY FROM THE RIGHT SUBCLAVIAN ARTERY

AUTHOR	AGE	SEX	ANOMALOUS ARTERIAL SUPPLY	CAUSE OF DEATH	ASSOCIATED ANOMALIES
Müller[58] (1927)	35 yr.		Artery size of brachial artery from right subclavian artery to hilum of right lung, where it divided into a plexus of vessels	Septicemia	Aortic stenosis, atresia of the right pulmonary artery; minor cardiac anomalies; small bilobed right lung
Menke[57] (1936)	60 yr.		Artery arose from right subclavian with thyrocervical trunk and coursed tortuously to right lung behind the main bronchus	Unknown	

TABLE VII. ABNORMAL ARTERY FROM THE INNOMINATE ARTERY TO THE HILUM OF RIGHT LUNG*

AUTHOR	AGE	SEX	ANOMALOUS ARTERY	CAUSE OF DEATH	ASSOCIATED ANOMALIES	OTHER FINDINGS
Breschet[61] (1826)	6 wk.		Artery connecting right subclavian and right pulmonary arteries near the origins of both vessels	Cardiac death	Cor biloculare, dextraposition of the aorta, patent ductus arteriosus	
Doering[62] (1914)	8 mo.	F	Artery from innominate artery to the upper part of the right hilum; it was 4 mm. in diameter, 12 mm. long, and had a constriction to 1 mm.	Bilateral bronchopneumonia	Aplastic right pulmonary artery, patent ductus arteriosus, patent foramen ovale	Left lung was larger than the right
Ambrus[59] (1936)	6 days	F	Artery 9.5 mm. in circumference from the innominate artery to the upper part of the right hilum	Hemorrhage into right lung	Functionally closed patent ductus arteriosus, aplastic right pulmonary artery, patent foramen ovale	Engorged right lung with blood in alveoli
Blalock[60] (1948)			Artery almost as large as the innominate artery from that vessel to the base of the heart; it became progressively smaller and had no lumen at its distal end	Operative finding		Cyanotic heart disease; the anomalous vessel was anastomosed to the right pulmonary artery

*The cases of Breschet and Blalock are included here for convenience.

Anomalous Pulmonary Blood Supply From the Right Subclavian Artery

The artery noted in Menke's[57] case was very tortuous. This feature may be confusing to the surgeon and hazardous to the patient.

Abnormal Artery From Innominate Artery to Hilum of Right Lung

All of these anomalies were observed in infants or children. The systemic artery coursed ultimately to the upper lobe of the right lung in 2 instances. In Blalock's[60] case the artery ended blindly at the base of the heart close to the right hilum, but it is included here because of its similarity to the other cases in this group and because of its surgical significance to be discussed later. In three of these cases there was associated atresia or hypoplasia of the pulmonary artery to the right lung.

At autopy Ambrus[59] found the alveoli of the right lung to be filled with red blood cells. He thought this was due to the excessively high blood pressure in the pulmonary vascular tree.

Arteries From the Aorta Proximal to the Origin of the Left Subclavian Which Course to the Hila of the Lungs

The ages of these patients vary widely. Systemic arteries supplied the lungs on both sides in 3 cases, the right side in 2, and the left side in 3 cases. More than one artery coursed to the right lung in 2 cases. A patent ductus arteriosus no doubt comprised the first portion of some of these vessels as they came off the aorta. In other instances it might be suspected that these anomalous arteries were hypertrophied bronchial arteries.

Cioni's case[47] is an interesting one, demonstrating dual circulation to the right upper lobe from both the greater and the lesser circuits for 69 years without apparent damaging effect on that lobe. In Sweet and White's case,[69] on the other hand, the upper lobe of the right lung was supplied only by an artery from the aorta, with eventual hemorrhage into the parenchyma at a relatively young age.

Serious cardiac anomalies were associated with the majority of these cases.

Case 4. Atresia of Right Pulmonary Artery and a Large Anomalous Artery to the Right Lung From the Ascending Aorta.—

C. A. D. (B. H. No. 887295, autopsy No. 7160), a 4-month-old female Negro infant, was admitted to the Babies Hospital, New York, N. Y. for the first time on Feb. 1, 1948, with the chief complaints of intermittent cyanosis, dyspnea, and fever.

Following a normal delivery and early postnatal period, she had two episodes of cyanosis, which were preceded by coughing and followed by vomiting which brought relief. On the morning of admission she had another similar attack which was the most severe.

She was well developed and well nourished. The skin was dusky gray. The respirations were rapid, and fine râles were heard at the bases of both lungs. The heart appeared to fill the entire left chest. Its rate was 180 and the rhythm was regular. The heart sounds were of good quality; a systolic murmur was heard over the precordium but was heard best in the third left interspace.

Laboratory examination revealed nothing unusual. Roentgenograms of the chest showed a greatly enlarged heart (Fig. 9). The vascular markings at its base were dis-

TABLE VIII. ARTERIES FROM THE AORTA PROXIMAL TO THE ORIGIN OF THE LEFT SUBCLAVIAN WHICH COURSE TO THE HILA OF THE LUNGS

AUTHOR	AGE	SEX	ANOMALOUS ARTERIAL SUPPLY	CAUSE OF DEATH	ASSOCIATED ANOMALIES	OTHER FINDINGS
Buchanan[63] (1864)	6½ mo.	F	Pulmonary arteries arose separately from the posterior aspect of the ascending aorta and coursed to each pulmonary hilum		Cor biloculare; atresia of the pulmonary artery (truncus arteriosus)	No cyanosis; normal lungs
Fraentzel[65] (1868)	25 yr.	F	From the ascending aorta an artery as thick as a finger coursed to the right hilum	Cardiac decompensation	Connection 12 mm. in diameter between pulmonary trunk and aorta; no right pulmonary artery	Ligamentum arteriosum
Kugel[67] (1931)	3 mo.		Large artery from aortic arch to right hilum; large artery from patent ductus arteriosus to the left lung	Cyanotic death	Atresia of the pulmonary orifice; patent ductus arteriosus; interventricular septal defect	
Ingalls[66] (1932)	8 wk.	F	Fair-sized artery from beginning of descending aorta to right lung; another artery from aorta near ductus arteriosus which bifurcated and sent a branch to each hilum	Bronchopneumonia	Atresia of proximal pulmonary artery; right aortic arch; cor biloculare; all pulmonary veins drained into superior vena cava	Another artery from descending aorta to right hilum; heart in good condition
Manca[68] (1933)	4½ mo.	F	Fair-sized artery from undersurface of aortic arch, which bifurcated, sending a branch to each hilum; no anastomoses with lesser circulation	Cyanotic since birth	Atresia of pulmonary orifice; right aortic arch; interventricular septal defect; hypoplasia of pulmonary artery	Ductus or ligamentum arteriosum not seen; 3 bronchial arteries to right lung; 2 arteries to left lung
Cioni[64] (1939)	69 yr.	M	Artery the size of the common iliac artery from proximal one-third of aortic arch coursed to right upper lobe dividing into several branches at hilum	Lobar pneumonia; chronic pericarditis		Normal pulmonary arteries to both lungs
Thomas[70] (1941)	3 mo.	F	Large artery from aorta at level of origin of left innominate artery to hilum of left lung	Cardiac and bronchopneumonia	Interauricular and interventricular septal defects; right aortic arch; absence of pulmonary valve; no normal left pulmonary artery	No ductus arteriosus or ligamentum; branches of aorta in order: right subclavian, right common carotid, left innominate
Sweet & White[69] (1944)	28 yr.	F	An artery larger than the descending aorta from the distal one-third of the aortic arch which divided, sending branches to the left lung	Operative finding; left upper lobe lobectomy	Atresia of the left pulmonary artery; evidence of cardiac anomaly	Hemorrhage into left upper lobe due to ruptured atheromatous branch of anomalous pulmonary artery

proportionately narrow, but the right hilar markings were more prominent than usual. There was an extensive irregular area of increased density in the right lung field. The electrocardiogram did not show right axis deviation as would be expected in a child of this age.

Following digitalization the child was considerably improved and she was discharged with the primary diagnosis of congenital heart disease with failure.

Second admission: After two days of general improvement at home, the infant again showed signs of acute respiratory embarrassment as well as fever of 102° F. She was re-admitted on Feb. 13, 1948. She now appeared acutely ill, dehydrated, and had gasping respirations. There were moist râles throughout both lung fields but especially at the bases. The heart sounds had not changed except that the systolic murmur had increased in intensity.

Fig. 9 (Case 4).—Demonstrating enlarged heart, diminished vascular shadows at base of heart, and irregular areas of increased density in right lung field.

The laboratory examination showed a slight increase in the white blood cell count with a normal differential. All other special examinations failed to reveal significant information. The patient's course became progressively worse and she died 18 hours after admission.

Autopsy findings: The body was that of a well-nourished, well-developed infant. The mucous membranes and nail beds were slightly cyanotic. The volume of the left lung was decreased by one-half due to the enlarged heart. The interlobar fissures were normal in number and location. On sectioning the lungs, scattered irregular hemorrhagic areas were noted throughout both lungs. There was frank consolidation of the right lower lobe. On microscopic study confluent lobular pneumonia was present in the upper and lower lobes of the right lung and the lower lobe of the left lung.

The greatly enlarged heart weighed 79.9 grams. There was equal dilatation and hypertrophy of both ventricles. Both auricles showed dilatation, but the right was twice as great as the left. Venae cavae and pulmonary veins entered the auricles at their usual sites. The coronary sinus was normal, but the foramen ovale was patent. The pulmonary valve measured 2.9 cm. and the aortic valve 2.2 cm. in diameter. The heart was otherwise normal.

The coronary arteries arose normally. One centimeter above the aortic valve a large vessel 2.7 cm. in diameter arose from the left side of the aorta (Fig. 10). This artery coursed behind the esophagus to enter the right lung.

The innominate and left subclavian arteries arose at their usual sites. The left common carotid artery branched from the innominate artery 1 cm. above its origin. The ligamentum arteriosum was normal in location.

The pulmonary artery was of normal caliber, measuring 2.5 cm. in diameter at its origin and 1.7 cm. in diameter beyond the dimple of the ligamentum. There was one branch to the left lung, but none to the right. Microscopic examination of the left pulmonary artery and the abnormal right pulmonary artery failed to show any difference in their mural structures.

The other autopsy findings were not contributory to this subject.

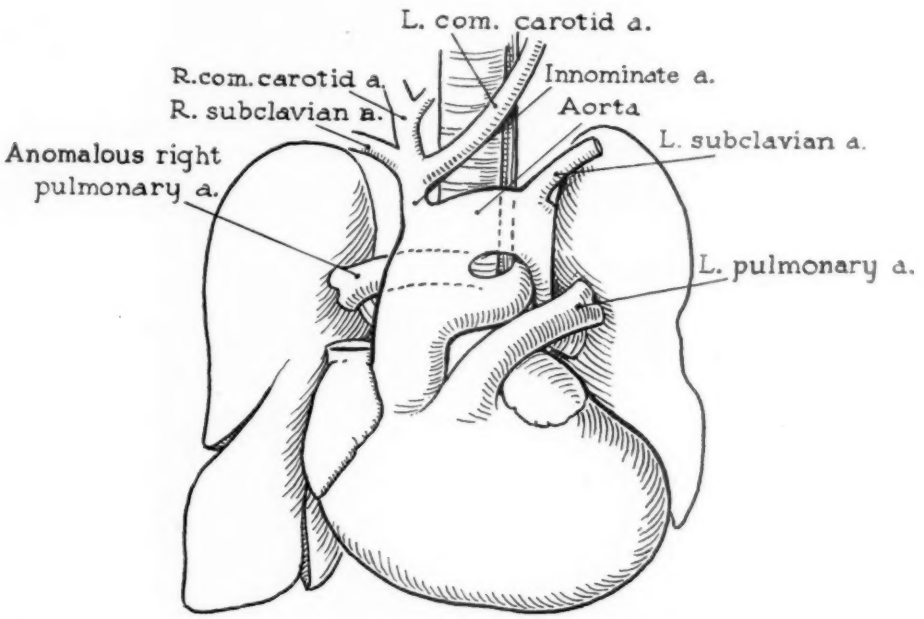

Fig. 10 (Case 4).—Anterior view illustrating heart and great vessels. (See text.)

Comment.—The gradual left cardiac enlargement was believed to be due to circulatory inefficiency caused by the short circuiting of a large volume of blood from the aorta through the right lung and back to the left auricle. As the left ventricle enlarged to encroach on and diminish the volume of the left lung, the pulmonary arterial blood pressure increased, with resultant right cardiac dilatation and hypertrophy. A vicious cardiorespiratory cycle ensued.

It was appreciated that the right lung with its anomalous circulation was functionless. With the gradual incapacitation of the functional left lung by the progressively enlarging heart, the diminished respiratory reserve was embarrassed to the point that it was incompatible with the life of the patient.

If the correct diagnosis could have been made early enough, it is probable that a right pneumonectomy or division of the anomalous pulmonary artery[86] would have benefited the infant.

Special Cases of Interest Not Falling Into the Preceding Classifications

The usual examples of patent ductus arteriosus have not been included in this collection, since they represent a common anomaly which has been discussed at length in recent years. An unusual type of Botallo's duct, however, is presented here because of its potential cause of dysphagia and respiratory symptoms had the patient lived longer. Combes[72] recorded a case of a stillborn child who died of an unknown cause (Table IX). The aorta left the heart normally, curved over the left main bronchus and behind the esophagus to descend

TABLE IX. ABNORMAL BLOOD FLOW FROM SUPERIOR VENA CAVA TO LEFT PULMONARY ARTERY

AUTHOR	AGE		ABNORMAL VASCULAR COMMUNICATION	CAUSE OF DEATH	ASSOCIATED ANOMALIES
Abbott[71] (1948)	14		A large vessel connected the superior vena cava and the left pulmonary artery with an aneurysm at the latter juncture	Removal of the vessel and aneurysm failed to improve the patient's condition and death ensued	Tetralogy of Fallot; right aortic arch
		LEFT SUBCLAVIAN ARTERY FROM PATENT DUCTUS ARTERIOSUS			
Combes & Christopherson[72] (1884)	Stillborn		Left subclavian artery arose from ductus arteriosus close to its aortic end, ascended behind the trachea, and coursed over the left first rib	Left aortic arch with right descending aorta; large patent ductus arteriosus between aorta opposite the right subclavian and the truncus pulmonalis, passed behind the esophagus	Branches of aorta in order: coronaries, right common carotid, left carotid, right subclavian

in the right chest. It gave off the coronary arteries, the right and left common carotid arteries, and the right subclavian artery in the sequence stated. The lesser circulation was normal except that a large patent ductus arteriosus left the truncus pulmonalis, and curved to the right behind the esophagus to enter the aorta opposite the origin of the right subclavian artery. From this ductus, close to the aorta, there arose a large artery which ascended behind the trachea and to the left, crossing the first rib to become the left subclavian artery. Should such a ductus be divided, it would be advisable to maintain continuity of the subclavian artery to obtain a more normal functional result.

Abbott's case[71] is the only example known to us of a major vascular communication between the pulmonary artery and the vena cava. This anomaly was found in a patient with a tetralogy of Fallot and a right-sided aortic arch. There was a large vessel connecting the superior vena cava and the first portion of the left pulmonary artery. It had an aneurysm at its pulmonic end. The anomalous vessel and aneurysm were removed with death of the patient shortly

thereafter. Abbott believed that blood probably had flowed from the systemic venous system to the pulmonary arterial system with functional benefit to the patient.

ANOMALIES OF THE PULMONARY VEINS

Abnormal connections between the pulmonary and systemic venous system and anomalous venous drainage to the right heart have been reviewed and discussed recently by Brody,[74] Brantigan,[73] and Taussig.[76]

Brody has written the most significant paper on this subject. He collected 100 cases from the literature and reported 2 of his own. His statistical analysis in this report merits comment. Of the 102 cases, 65 demonstrated only partial venous drainage from the lungs to the right heart. This was found to be compatible with longevity in that 35 patients were adults. There were probably more that were in this age group since the age was not reported in 19 cases. One individual lived to be 86 years old. Abnormal pulmonary venous drainage was twice as common on the right as on the left side. Pulmonary veins draining into the superior vena cava were observed 29 times, into the right atrium 18 times, and into the left innominate vein in 14 instances. Less common drainage sites were the coronary sinus, inferior vena cava, azygos, and the left subclavian veins.

There were 37 cases in which the pulmonary venous drainage emptied completely into the right atrium or one of its tributaries. Of these, only 8 patients lived beyond 6 months of age, and the oldest was 27 years old. All of the older patients in this group had intracardiac shunts, usually a patent foramen ovale, which permitted mixing of the venous blood. Other great vessel and cardiac anomalies may be present to complicate the clinical picture further.

Brody[74] noted that in 25 patients in whom 75 per cent or more of the venous blood from the lungs returned to the left atrium, all but 2 reached adulthood. These died from other causes. He concluded that a heart without an interauricular septal defect can function adequately if no more than 50 per cent of the pulmonary blood returns to the right heart.

Taussig[76] has described fully the clinical findings associated with these anomalies. Concerning the treatment of complete venous drainage to the right heart, she stated that anatomic correction can be performed only when we are able to transplant the pulmonary veins into the left auricle. She made no mention of surgical therapy for the partial abnormal pulmonary drainage in that she considered that this will not cause significant symptoms and that it is compatible with life. Brantigan,[73] however, believed that if there is partial pulmonary drainage to the right heart, removal of the abnormally drained lobe will decrease the burden on the right heart.

If, for any reason, a surgeon performs a thoracotomy and observes pulmonary venous drainage coursing ultimately both to the right and left hearts, ligation and division of the vein going to the right, and reanastomosis of its distal end to a vein going to the left heart, would accomplish normal anatomic restoration of the venous return. The recent work of Gross and associates[75]

on the use of arterial homografts for bridging defects of the great vessels about the heart should be considered in the treatment of these anomalies by corrective surgery.

Case 5. Anomalous Vein Draining the Right Lung.—

M. P. (L. H. H.), a 26-year-old white woman, was admitted to the Lenox Hill Hospital because of an abnormal shadow in a roentgenogram of the chest which was taken because of an unexplained weight loss. She was said to have had asthma since childhood, but no other pulmonary symptoms.

The chest film showed a band of increased density measuring 4 cm. in its greatest diameter in the mediobasal quadrant of the right lung field adjacent and parallel to the right heart border (Fig. 11). In the lateral projection this area of density was situated in the mid-thoracic region.

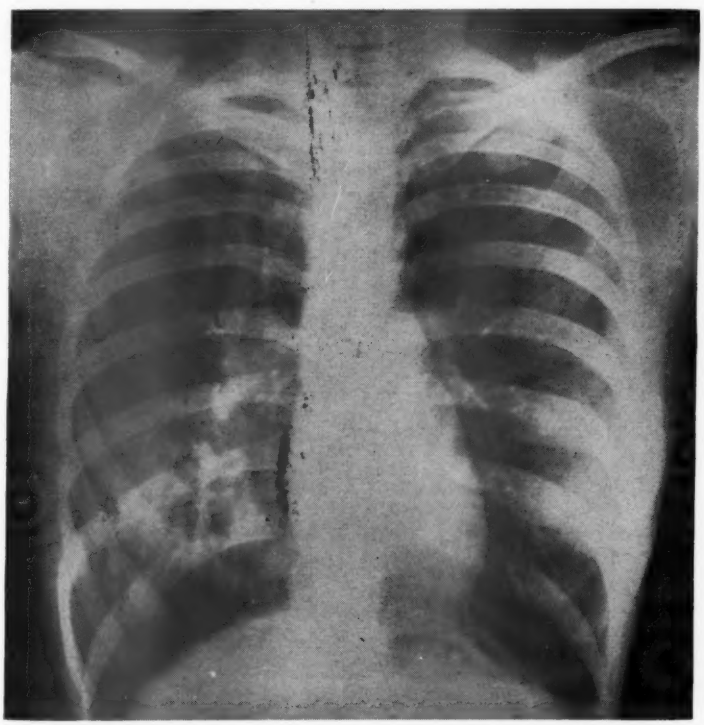

Fig. 11 (Case 5).—Roentgenogram of chest showing diminished vascular shadows in the upper one-half of the right lung field and abnormal linear shadows in the lower one-half of the right chest.

Bronchoscopy revealed an anomalous arrangement of the right bronchial tree. Angiocardiography proved that the shadow in the right lung field was an aberrant pulmonary vein apparently draining the entire right lung (Fig. 12). There was no evidence of an arteriovenous aneurysm.

Comment.—The health of this patient so far coincides with the statements of Brody.[74] If she develops signs of right heart failure, however, a right pneumonectomy may be advisable in accordance with the suggestion of Brantigan.[73] Cardiac catheterization with mensuration of the right auricular and ventricular pressures might give information in prognosticating this patient's future.

EMBRYOLOGY

There is no undisputed embryologic concept that can explain fully all of the anomalies in question. Investigators who have studied very early embryos agree that the arterial system develops from a capillary plexus.[81] Such an arrangement could explain theoretically any abnormal vascular communication or pulmonary artery in a term fetus. Unfortunately, as the later embryonic stages have been studied, none of the appropriate connections have been seen to persist, and most of the authoritative observations seem to militate against a satisfactory theory.

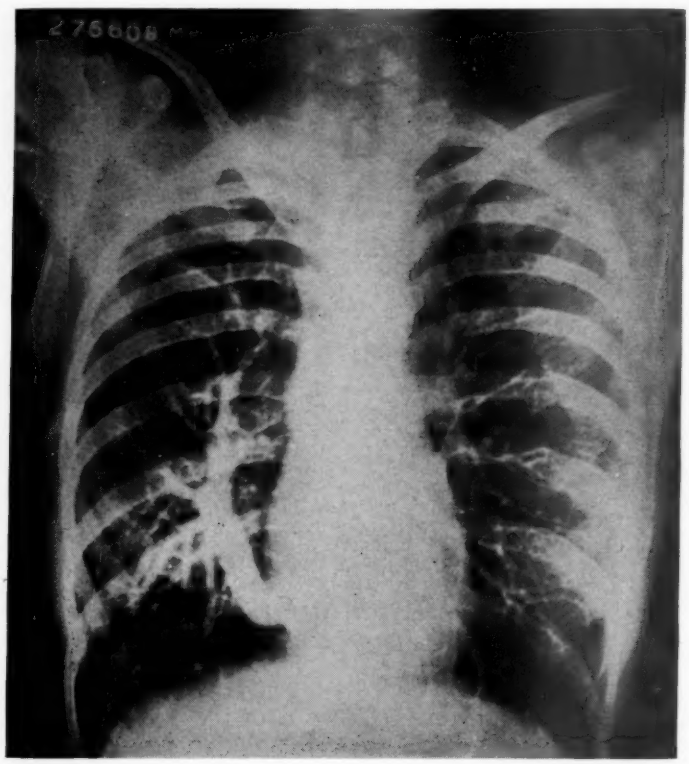

Fig. 12 (Case 5).—Angiocardiogram demonstrating large aberrant pulmonary vein on the right. Most of the Diodrast has already entered the left side of the heart and aorta. The carotid arteries are delineated. (See text.)

Between the 3 and 4 mm. stages a ventral ridge appears in the floor of the esophagus just caudal to the pharyngeal pouches; this is the earliest indication of the future respiratory organs. The primordial lung buds and trachea become separated from the gut up to the fourth brachial arch.[77] It is at this stage of development that divergent opinions on the circulation are obvious.

Brown[78] and Huntington[80] believed that an independent capillary plexus develops about the intestinal tract which anastomoses with the more prominent systemic circulation. A portion of this splanchnic plexus becomes intimate with the pulmonary anlage as it is formed. With the further development of the pulmonary plexus, communications are made cephalad with capillaries around

the primitive aortas. Caudad it anastomoses with the splanchnic plexus at the level where the hepatic sinusoids are forming. According to these authors, temporary direct communications between the pulmonary and systemic vascular systems, both superiorly and inferiorly, are therefore normal in the primitive embryo.

Although this thesis could explain adequately the anomalies in question, Congdon[79] disagreed with it. His reasons for disagreement are based on observations of his own as well as those of other embryologists. In his preparations he showed that the pulmonary plexus was derived from outgrowths of the aortic sac and the primitive dorsal aortas just below the fourth aortic arches. He noted capillary connections between the pulmonary and splanchnic networks at the level of the fourth aortic arches, but he did not agree with Brown or Huntington that these plexuses developed independently from the rest of the vascular system. It can be concluded from his observations that he did not consider the capillary anastomoses to be potential primordia of abnormal pulmonary circulations. From this authoritative monograph it is difficult to find a rationale for the anomalous arteries under discussion.

The embryology of the venous pulmonary system has been reviewed by McManus,[82] and more recently by Brantigan.[73]

DISCUSSION

An attempt has been made to survey the literature on abnormal pulmonary blood flow from the systemic circulation. We have tried to consider only cases of surgical significance, realizing that the criteria for selection is changing constantly. Therefore, certain types of cases have been omitted which may be worthy of consideration in the near future.

The groups of anomalies presented first have concerned anomalous vessels arising from the descending aorta. These vessels are the most peripheral in origin and they usually are not associated with the serious cardiac anomalies that are seen with aberrant pulmonary arteries or communications which arise closer to the heart. Accordingly, attention has been directed first to the more uniform cases which permit the formulation of generalities. Later the more complex cases were presented which are essentially unique in themselves. When the origin of the anomalous pulmonary vessel arose from the systemic arterial system above the level of the lung roots, there were associated significant anomalies of the heart or great vessels in 75 per cent of the cases. Such concomitant abnormalities were observed in only 16 per cent of cases demonstrating abnormal pulmonary vessels arising below this level.

Some of the systemic arteries which coursed along a main bronchus and finally became lost in lung tissue have been thought to be bronchial arteries. Cioni[64] considered his cases to be examples of heterotopic bronchial arteries. Usually the bronchial arteries arise at the level of the fifth or sixth thoracic vertebrae. Most commonly the left lung has 2 such vessels and the right lung has one.[90] Occasionally such arteries have been seen to arise from the internal mammary and intercostal arteries.[96] Many of the anomalies presented here

may function as bronchial arteries, but it is difficult to explain their location embryologically. Usually they were found to have larger calibers than the hypertrophied collateral vessels associated with pulmonary stenosis or atresia. In some instances where authors were fully justified in suspecting these abnormal vessels to be bronchial, normal bronchial arteries were also identified arising from their usual locations.

In discussing the abnormal arteries to the lower lobes it is interesting to note the gradation of associated anomalies of the pulmonary lobes. An aberrant lobe has no bronchial connection with the remainder of the tracheobronchial tree, and its blood supply is usually totally independent from the lesser circulation. The more normal cases listed here are those which have the usual bronchial patterns and communications although the arterial supply arises completely or partially from the greater circulation. Eppinger[48] reported an anomaly which has features of both these extremes. The right lung was bilobed but smaller than usual. The upper lobe bronchus had ultimate egress to the trachea but the lower lobe bronchus had no proximal communication. Both lobes were supplied by small branches of the right pulmonary artery and arteries from below the diaphragm.

Some of the aberrant lobes with their imperforate or atretic bronchi had undergone cystic changes. Congenital cysts were also noted in lower lobes which had normal bronchial communication, but which were supplied in whole or in part by systemic arteries. Since most of the aberrant lobes have been observed in infancy, it is interesting to speculate as to whether they atrophy in later years or become bronchiogenic cysts.

These cases have been reviewed to make the clinician and especially the surgeon more aware of such vascular anomalies. By arranging them in anatomic groups and studying their associated findings, an attempt will be made to establish criteria for making preoperative diagnoses of their presence.

Douglass[35] noted that when a thin-walled cyst with bronchiogenic characteristics is present in the base of the lower lobe, it is probably congenital in origin. He thought these cysts were accompanied frequently by anomalous pulmonary arteries and he described several demonstrative cases.

From the data collected there are other findings which should arouse suspicion of vascular aberrations. In a significant number of cases, anomalous bronchial arrangements have been observed. They may or may not be associated with abnormal lobes or fissures of the involved lungs. Less prominent hilar shadows by roentgenography may be corroborative evidence that the major portion of the pulmonary blood supply is arising from an unusual location (Case 5). When the diagnosis of congenital heart disease has been made, especially in the presence of chronic cyanosis, the possibility of aberrant collateral circulation should be entertained.

In order to determine the preceding diagnostic features, the following procedures should be performed. Roentgenograms should be taken both in the posteroanterior and lateral projections to note the configuration of the hilar shadows and the presence of thin-walled cysts or discrete opacities in the pul-

monary fields. Sometimes the number and position of the interlobar fissures can be ascertained. Bronchoscopy should then be done to determine unusual locations of the secondary and tertiary bronchial orifices. Bronchography will be helpful in noting abnormal or incomplete bronchial distribution. With the recent advances in angiocardiography, it is now possible to examine newborn infants by this method.[89, 95] Since many films can be taken during one examination, it is possible to outline both the cardiac chambers and many of the vessels arising from the descending thoracic aorta. Visualization of the aorta and its branches can be performed clearly and accurately by retrograde arterial injection of Diodrast.[87, 88, 94]

Once the diagnosis of an aberrant pulmonary artery or communication has been established, as precise physiologic knowledge as is feasible should be obtained of the heart and great vessels. Cardiac catheterization may aid in determining the direction and amount of blood flow through an anomalous communication as well as the intracardiac hemodynamics.[91, 92] Information obtained by this method may prevent the operative mistakes made in our case (Case 1) and in that of Abbott.[71] In these instances anomalous vessels, which were of functional significance to the patient, were divided.

Bronchospirometry may aid in evaluating more completely the functional role of the pulmonary parenchyma that is supplied by a systemic artery. If a lung or a lobe of a lung is functionless, the question of pulmonary resection arises, especially if this abnormality is physiologically harmful. It is of more than academic importance to examine by this method a patient who has pulmonary venous drainage emptying into the right heart or its tributaries since Brantigan believed that such an anomaly may cause a right heart strain.[73]

Once a surgeon has entered the thorax, there are signs which should make him suspect abnormal pulmonary circulation. The discovery of an aberrant lobe needs no comment. If he is operating for suppurative disease of the lung and discovers few or no associated pleural adhesions, he should consider the presence of a pulmonary anomaly with possible concomitant abnormal vascular supply. Unusual pulmonary lobes and interlobar fissures not shown by roentgenography may now be demonstrable. The pulmonary artery should be identified to see if its caliber is normal. If the pulmonary artery or one of its main branches is smaller than usual, there may be an anomalous vessel supplying the lung.

It is difficult to determine the prognosis in all of these cases, but a few generalizations can be made. When the heart and pulmonary arteries are normal and there is a direct anastomosis between the greater and lesser circulations, the prognosis probably should be considered the same as that of patent ductus arteriosus.

The prognosis is somewhat nebulous if the pulmonary artery is normal and there is also a large arterial vessel from the systemic circulation which functions like a bronchial artery. It is necessary to review previous observations to postulate a prognosis. Berry and Daly[83] injected the bronchial arteries of stillborn infants and noted that the perfused substance emptied into the pulmonary

veins and arteries at an approximate ratio of 3:1. In normal lungs the communication between all these vascular systems are through capillaries. Wood and Miller[96] observed, however, that sometimes in certain pathologic conditions such as silicotuberculosis and congenital cystic disease of the lungs, there were large bronchopulmonary vascular anastomoses. Some of these patients died of right heart failure which was ascribed in part to pulmonary hypertension. From these observations it is conceivable that such an outcome is possible in any case where a lung or a part thereof is supplied by arteries from both the greater and lesser circulations. Cioni's second patient,[64] however, reached the age of 69 years without suffering from right cardiac decompensation.

In order for an individual to survive with a markedly hypoplastic or absent pulmonary artery, hypertrophied arteries from the greater circulation are necessary. These enlarged vessels may be normal or aberrant. To surgeons operating for the correction of pulmonic stenosis, increased collateral circulation to the lungs is a common finding. Miller[93] reported an extreme example of the failure to develop an adequate compensatory circulation. In his case 1 lung had neither a pulmonary artery nor a complementary systemic artery. Gangrene of the lung occurred, leading to spontaneous perforation of the visceral pleura and death by tension pneumothorax.

Müller's case[58] is of interest in that the patient lived to the age of 35 and apparently died of causes other than her congenital anomaly. Aortic stenosis and atresia of the right pulmonary artery were present. A vessel the size of the brachial artery arose from the right subclavian artery and coursed to the right main bronchus, where it divided into a plexus of vessels. There was no evidence of pulmonary hypertension or congestion.

There are, however, other cases which exemplify several untoward sequelae. Ambrus'[59] infant demonstrated an aplastic right pulmonary artery and a large vessel arising from the innominate artery which entered the upper part of the hilum of the right lung. At autopsy this lung was engorged and there were red blood cells in the alveoli. The pathologic process was ascribed to a pulmonary hypertension which was sufficient to cause a diapedesis of red corpuscles through the capillaries of the lung or an actual rupture of these vessels.

Sweet and White's[69] patient was 28 years of age when she came to them for treatment. She had been relatively asymptomatic until shortly before her admission to the hospital, when she began to complain of intermittent hemoptysis. At operation no normal pulmonary artery on the left was found. There was, however, a large artery arising from the distal end of the aortic arch which branched to supply the lobes of the left lung. The upper lobe of the left lung was removed. Study of the specimen showed hemorrhage into the lung substance which apparently resulted from rupture of an atheromatous branch of this anomalous pulmonary vessel. This case raised the question as to whether an abnormal artery to the lung coming from the greater circulation is more susceptible to degenerative changes than normal arteries elsewhere in the body.

The studies of Bloomer and associates[86] related to this subject are significant. They ligated a main branch of a dog's pulmonary artery and subse-

quently studied the cardiopulmonary function. A few weeks after operation they noted a marked increase in the bronchial arterial circulation. Blood from these vessels perfused the alveolar capillaries via precapillary anastomoses and the transfer of gases was efficiently accomplished. This abnormal blood flow, however, increased the burden of the left heart, whose output became one-third greater than the right. Case 4 is an extreme example of the result of such a vascular arrangement due to the tremendous size of the aberrant pulmonary artery.

From the functional standpoint, the efficiency of collateral circulation to the lung in the presence or absence of cyanosis has not been evaluated fully. By injection studies of specimens removed post mortem or following excisional surgery, intrapulmonary communications between the arteries of the greater and lesser circulation have been demonstrated.[42, 50] In the living, however, the resistance to blood flow from one system to the other has not been measured in patients with many of the pathologic features presented here. This is due partially to technical difficulties peculiar to these patients and at times to the inability to diagnose completely the pathologic hemodynamics before operation or death. The work of Bing[84, 85] and his colleagues, however, has contributed much to the understanding of this problem. In the future valuable information may be obtained through more accurate preoperative diagnosis, cardiac catheterization, bronchospirometry, and postoperative injection studies.

With further evaluation of these cases several points of surgical significance may be stressed. If these aberrant arteries are not recognized or handled carefully, death may ensue due to fatal hemorrhage. Operative fatalities have been reported to illustrate this point.[35, 36] These arteries are often tortuous, delicate, and occasionally surrounded by adhesions. Arce[45] believed that all sizeable adhesions should be divided between clamps and ligated to avoid possible uncontrollable bleeding. If these vessels are divided without the aid of hemostats, they bleed briskly due to their high intraluminal blood pressures and they tend to retract beyond the reach of the operator.

In cases of infected congenital cysts of the lower lobe, a surgeon should beware of pneumonotomy. In the first place, pleural adhesions in the region of the cyst are often absent and our case (Case 3) demonstrates a failure of producing adhesions by the two-stage drainage method. But a more important feature is the pericavitary vascularization by arteries having high intraluminal blood pressures. As demonstrated by the injection study of the specimen removed from Case 3 (Fig. 8), the vessels surrounding the cavity are large and very numerous. It is easily conceivable how uncontrollable hemorrhage might result from open drainage of such a pulmonary cyst.

Case 5 illustrates that a needless operation may be prevented if diagnostic acumen is acute. Although the patient was referred to one of us for surgery, the roentgenographic signs led to the suspicion that a pulmonary vascular anomaly was present. Accordingly, special studies were performed and operation was withheld. Needless to say, this individual will have to be followed

closely in the future for untoward signs and symptoms, but nevertheless there did not seem to be sufficient reason to operate on the patient at the time of examination.

In recent years the technique of segmental resection of a pulmonary lobe has become popularized. Thoracic surgeons are striving more and more to preserve as much healthy lung parenchyma as possible in extirpative surgery of benign lesions. If examples of some of the cases just presented are to be encountered, a word of caution on segmental resection is advisable. By injection studies, Paul[42] noted in his case that the aberrant artery which supplied the mediobasal portion of the lower lobe of the left lung anastomosed profusely with the pulmonary arteries from the lesser circulation in the adjacent lung parenchyma. Maugars,[50] by a similar method, also showed anastomotic connections between the two vascular systems. From these findings it would seem likely that a segmental resection of a diseased area, which is supplied by an aberrant artery, might be fraught with danger, or at least would present significant technical difficulties.

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Book Reviews

Books Received

The receipt of books is acknowledged in this section and this treatment must be regarded as sufficient acknowledgment of the courtesy of the senders. Selections will be made for more extensive review dictated by the interest of our readers as space permits.

SURGERY ORTHODOX AND HETERODOX. By Sir William Heneage Ogilvie, K.B.E., DM., M.Ch., F.R.C.S., Hon. F.R.C.S., Canada, Hon. F.R.A.C.S., Member of the Council, Royal College of Surgeons; Surgeon, Guy's Hospital and Royal Masonic Hospital. Hard. Price $4.00. Pp. 241, with 30 illustrations. Springfield, Ill., 1948, Charles C Thomas.

VISUAL ANATOMY: HEAD AND NECK. By Sydney M. Friedman, M.D., Professor of Anatomy, University of British Columbia, Vancouver, Canada. Hard. Price $6.50. Pp. 232, with 93 illustrations. Springfield, Ill., 1950, Charles C Thomas, Publisher.

INDICATIONS FOR AND RESULTS OF SPLENECTOMY. By Frederick A. Coller, M.D., Alexander Blain III, M.D., and Gould Andrews, M.D., Department of Surgery and Medicine, University of Michigan Medical School, Ann Arbor, Mich. Hard. Price $2.25. Pp. 100, with 16 illustrations. Springfield, Ill., 1950, Charles C Thomas, Publisher.

SKULL FRACTURES AND BRAIN INJURIES. By Harry E. Mock, M.D., Consulting Surgeon, St. Luke's Hospital, Chicago, Ill.; Associate Professor Emeritus of Surgery, Northwestern University Medical School, Chicago, Ill. Hard. Price $13.50. Pp. 806, with 116 illustrations. Baltimore, Md., 1950, Williams & Wilkins Company.

HIRNDURCHBLUTUNGS—STORUNGEN. By Von A. Brobeil. Hard. Price 45 francs. Pp. 264, with 84 illustrations. Stuttgart, 1950, Georg Thieme verlag.

AN OUTLINE OF UROLOGY. By C. D. Creevy, M.D., Professor of Surgery, Director Division of Urology, University of Minnesota Hospital, Minneapolis, Minn. Soft. Price $3.75. Minneapolis, Minn., 1950, Burgess Publishing Company.

YOUR PROSTATE GLAND. By Reed M. Nesbit, M.D., Professor of Surgery, University of Michigan Medical School; Chief, Section on Urology, University Hospital, Ann Arbor, Mich. Hard. Price $2.00. Pp. 50, with few illustrations. Springfield, Ill., 1950, Charles C Thomas, Publisher.

BRITISH SURGICAL PRACTICE. By Sir Ernest Rock Carling, F.R.C.S., F.R.C.P., and J. Paterson Ross, M.S., F.R.C.S. Vol. 8. Hard. Price $13.20. Pp. 597, with 264 illustrations. St. Louis, Mo., 1950, The C. V. Mosby Company.

VESALIUS (THE ILLUSTRATIONS FROM HIS WORK). By Saunders and O'Malley. Hard. Price $10.00. Pp. 248, with 96 illustrations. Cleveland and New York, 1950, World Publishing Company.

ATLAS OF HISTOLOGIC DIAGNOSIS IN SURGICAL PATHOLOGY. By Dr. Karl T. Neubuerger, Professor of Pathology, University of Colorado School of Medicine, Denver, Colo. Hard. Price $11.00. Pp. 460, with 880 illustrations. Baltimore, Md., 1950, Williams & Wilkins Company.

HAROFE HAIVRI. The Hebrew Medical Journal. Edited by Moses Einhorn, M.D. New York, 1950.